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BONE HEALTH IN OLDER ADULTS WITH AN INTELLECTUAL DISABILITY



Trinity College Dublin

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A thesis submitted to the University of Dublin, Trinity College, for the Degree of Doctor of Philosophy

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Date: 14/10/2016

DEDICATION

In loving memory of two of the strongest and most beautiful women I have had the privilege to ever know, Elizabeth and Evelyn, my mum and mum-in-law. I dedicate this thesis to you both in admiration of the work you did, the life you gave and the lessons I have learned. Thank you both.

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PUBLICATIONS, AWARDS AND CONFERENCE PRESENTATIONS

Publications

Burke, EA., McCallion, P., Carroll, R Walsh, JB and McCarron, M (2016). An exploration of the bone health of older adults with an intellectual disability in Ireland. *Journal of Intellectual Disability Research* doi: 10.1111/jir.12273.

Burke, EA., McCallion, P., Walsh, JB and McCarron, M (Under reviewed) Osteoporosis and people with Down syndrome; a preliminary descriptive examination of the Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing Wave 1 results. Submitted to the special edition, *Journal of Policy and Practice in Intellectual Disability.*

Academic Awards and Prizes

March 2013; School of Nursing Postgraduate Research Studentship, Trinity College Dublin 'An exploration of the bone health of adults with an intellectual disability'

September 2013; **Burke**, EA., McCarron, M. and McCallion, P *'Osteoporosis and people with Down syndrome; A preliminary descriptive examination of the IDS-TILDA Wave 1'* Winning juried SIRGAIDD Award for best student poster at the Boston roundtable of the IASSIDD special interest group on ageing with an intellectual disability.

Keynote Address

Burke, EA. (2015) The lives and needs of older people with learning Disabilities in Ireland. BILD Annual International Conference, Growing older with a learning disability, Birmingham. December 4th 2015.

Burke EA. (2015) Unlocking the determinants of ageing and bone health among adults with an intellectual disability – Results from IDS-TILDA. Disability Research Seminar Series November 3rd 2015

Burke EA. (2015) Health, well-being and community participation: IDS-TILDA. Seminar delivered to Camphill Community National Conference, Kilkenny.

Oral Presentations

Burke, EA, McCallion P, Carroll, R, Walsh JB. and McCarron M (2016) Bone status in people with intellectual disability. 2nd Annual Multidisciplinary Study Day in Metabolic Bone Biochemistry and Medicine, Clinical Biochemistry, School of Medicine, Trinity Centre for Health Sciences, Tallaght Hospital.

Burke, EA., McCallion, P., Walsh, JB. and McCarron, M. (2015) Examining the bone health of older adults with ID. Irish Gerontological Society PhD Conference. Education Center, Our Lady's Hospice, Harold's Cross, Dublin.

Burke, E., McCallion, P., Walsh, J., & McCarron , M. (2015). *Engaging older adults with an intellectual disability in health assessments; Health Assessment Development Wave 2 of the Intellectual*

Disability Supplement to the Irish Longitudinal Study on Ageing (IDS-TILDA). University of Dublin, Trinity College Dublin. Unpublished.

Burke, E., McCallion, P., Walsh, J., & McCarron, M. Structured Feedback from Older Adults with an Intellectual Disability on their Experience of Attending for a Health Assessments A Feasibility Study. *Age, 40*(49), 3.

Burke, EA., McCarron, M., Walsh, J.B. and McCallion, P. (2014). Developing a baseline picture of the bone health status of older adults with an ID in Ireland. 4th IASSID (International Association for the Scientific Study of Intellectual and Developmental Disabilities), European Congress, Pathways to Inclusion University of Vienna, Austria.

Burke, EA., McCarron, M., Walsh, J.B. and McCallion, P. (2014). Supporting older adults with ID to engage in health screening: lessons learned from the Intellectual Disability Supplement to the Irish Longitudinal Study on Ageing. 4th IASSID (International Association for the Scientific Study of Intellectual and Developmental Disabilities), European Congress, Pathways to Inclusion University of Vienna, Austria.

Burke, EA., McCallion, P. and McCarron, M. (2013) The Silent Thief of Bone: Developing a baseline picture of the bone health of adults with an intellectual disability in Ireland. 14th Interdisciplinary Research Conference Health, Wellbeing and Innovation: recent advances in research, practice and education, Dublin, Ireland.

Poster Presentations

Burke EA., McCallion P., Walsh, JB. & McCarron M (2015). The Bone Status of Adults with Intellectual Disability, Results from the Second Wave of IDS-TILDA. Unlocking the Demographic Dividend. The International Association of Gerontology and Geriatrics European Region Congress 2015, Dublin, Ireland.

Burke EA., McCarron M., Walsh, JB. and McCallion P. (2015).The Feasibility of Utilising Quantitative Ultrasound as a Method of Objectively Measuring Bone Status among Older Adults with Intellectual Disability. Unlocking the Demographic Dividend. The International Association of Gerontology and Geriatrics European Region Congress 2015, Dublin, Ireland.

Burke, EA., Carroll, R., Walsh, JB., McCallion, P. and McCarron, M. (2015) Examining the risk factors for bone fragility among of older adults with an intellectual disability in Ireland. WCO-IOF-ESCEO Milan 2015.

Burke, EA., McCarron, M., Walsh, J.B. and McCallion, P. (2014). Structured feedback from older adults with an intellectual disability on their experience of attending for a health assessment: Results from a pilot study. 4th IASSID (International Association for the Scientific Study of Intellectual and Developmental Disabilities), European Congress, Pathways to Inclusion University of Vienna, Austria.

Burke, EA., McCallion, P., Walsh, JB. and McCarron, M. (2014). Developing a baseline picture of the bone health status of older adults with an ID in Ireland. World Congress on Osteoporosis, Osteoarthritis and Musculoskeletal Diseases (WCO-IOF ESCEO), Seville, Spain.

Burke, EA., McCallion, P., & McCarron, M. (2013)Osteoporosis and people with Down syndrome; a preliminary descriptive examination of the IDS-TILDA Wave 1 results, . *Annual Roundtable - Special Interest Group on Aging with an Intellectual Disability, International Association for the Scientific Study of Intellectual & Developmental Disabilities, Boston, MA, September 2013*, (Winning entry).

Reports and Book Chapters

Burke, EA., McCallion, P. and McCarron M. (Eds) (2015) Advancing Years, Different Challenges: Wave 2 IDS-TILDA Findings on the ageing of people with an intellectual disability. Dublin: School of Nursing & Midwifery, Trinity College Dublin.

McCarron, M., Swinburne, J., **Burke**, EA., McGlinchey, E., Mulryan, N., Andrews, V., Foran, S. and McCallion P. (2011) Growing Older with an Intellectual Disability in Ireland, First results from the Intellectual Disability Supplement to the Irish Longitudinal Study on Ageing. School of Nursing & Midwifery, Trinity College Dublin.

McCallion, P. Swinburne, J., **Burke**, EA., McGlinchey, E., & McCarron, M. (2013). Understanding the similarities and differences in aging with an intellectual disability: Linking Irish general population and intellectual disability datasets. In R. Urbano (Ed.). *Using Secondary Datasets to Understand Persons with Developmental Disabilities and their Families* (IRRDD-45 pp.206-233). NY: Academic Press.

Accessible Publications

Burke, EA. (2015) Investigating osteoporosis among adults with intellectual disability. Accessed at: https://www.youtube.com/watch?v=FdfQkcy7Z-M, Innovation Academy Trinity College Dublin.

McGlinchey, E., McCausland, D., **Burke**, EA., Carroll, R., O'Donovan, MA., Mulryan, N., Shivers, C., O'Dwyer, M., McCallion P and McCarron, M. (2015) Advancing Years, Different Challenges: Wave 2 IDS-TILDA Findings on the ageing of people with an intellectual disability. An Accessible Report, Dublin: School of Nursing & Midwifery, Trinity College Dublin.

McCarron, M., McGlinchey, E., **Burke** EA. & McCallion P. McGlinchey E. & **Burke E.** Producers (2012) *Scenes from Our Lives* [DVD].

McCarron, M., Swinburne, J., **Burke**, EA., McGlinchey, E., Mulryan, N., Andrews, V., Foran, S. and McCallion P. (2011) Growing Older with an Intellectual Disability in Ireland, First results from the Intellectual Disability Supplement to the Irish Longitudinal Study on Ageing –An Accessible Report.

Seminars

Burke EA., (2015). Results from the IDS-TILDA study Wave 2, Determinants of health among adults with an intellectual disability –Speech and Language Department Seminar, November 24th 2015

Burke, EA. (2014). Health concerns for adults with intellectual disability as they grow older. 4th Year RNID Student Nurses: Contemporary Issues in Intellectual Disability Nursing, Trinity College, Dublin, Ireland.

Burke, EA. (2014). The Intellectual Disability Supplement to the Irish Longitudinal study on ageing: Research among older adults with ID. 4th Year RNID Student Nurses: Contemporary Issues in Intellectual Disability Nursing, Trinity College, Dublin, Ireland.

Burke, EA. (2014). The changing health profile of adults with ID: Results from Wave 2 IDS-TILDA. National Disability Authority (NDA), Dublin, Ireland.

Burke EA., McCarron and McCallion (2014) I'm not ready yet, what people with an ID say about growing older and retirement in Ireland. Royal College of Surgeons Ireland. June 2014.

Burke. EA., McCallion, P. and McCarron, M. (2014). Bone health and people with ID. A Comprehensive Approach to Health and Health Care for People with Intellectual Disabilities as they Age, IASSIDD Academy on Education, Teaching, and Research. 4th IASSID (International Association for the Scientific Study of Intellectual and Developmental Disabilities) European Congress, University of Vienna, Austria.

Burke, EA., McCarron, M. and McCallion, P. (2014). A preliminary examination of the bone health of older adults with intellectual disability in Ireland: IDS-TILDA Wave 1. Tung Wah Group, Taiwan International Exchange, Dublin, Ireland.

Burke EA., McCarron, M., McGlinchey, E., McCallion P. (2013) Exploring a Picture of Ageing for People with an Intellectual Disability in Ireland Research and Practice Seminar St Michael's House Service Dublin.

Burke, EA., McCarron, M., Swinburne, J., McGlinchey, E., Mulryan, N., Andrews, V., Foran, S. and McCallion P. (2011). Social Connectedness and Social Participation of Older adults with an Intellectual Disability in Ireland 2011: First results from IDS-TILDA, Irish Nurses and Midwives Organisation Education Day, Dublin, Ireland, December 2011.

ABSTRACT

Introduction

Intellectual disability (ID) is characterised by significant limitations of both adaptive behaviour and intellectual functioning. It is a prescribed term to describe a common understanding of what is being discussed or studied and it is not for the purposes of labelling or describing people. Currently the life expectancy of people with ID is increasing. However, the Health Service Executive (HSE 2005) cautions that an increased older population could give rise to a significant increase in chronic diseases, one of which is osteoporosis. It has been reported that people with ID are two and half times more likely to present with adverse health conditions than the general population. Osteoporosis is strongly associated with ageing and as the older population of people with ID increase so too does their susceptibility to osteoporosis. Osteoporosis is a systematic skeletal disease characterised by microarchitectural deterioration of the bone with consequent increased vulnerability to fragility fracture. As an insidious condition it is very often only diagnosed post first clinical fracture.

Background & Justification

Bone health underpins healthy ageing and is a major determinant of successful ageing. People with ID present with increased risk of low bone mineral density especially those with, for example Down syndrome. Moreover it is increasingly recognised and of urgent concern that osteoporosis continues to be under diagnosed and undertreated in the older population of people with ID. Despite an acknowledged increased loading toward osteoporosis, people with ID are less likely to have bone health checks or routine screening. Considering this and the impact of fracture on quality of life, investigating bone health among people with ID is timely and required.

Aim

The aim of this study is to identify the prevalence of osteoporosis and osteopenia among adults with an ID in Ireland, to examine the predominant predictors of poor bone health among this cohort and to establish a robust method of engaging all people with ID in health assessment particularly bone health assessment.

Methods and materials

This study was conducted within a larger longitudinal study on ageing and ID namely, *The Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing* (IDS-TILDA). This is comparable with the Irish Longitudinal Study on Ageing (TILDA). In the IDS-TILDA study people with all levels of ID, over the age of 40 years were randomly selected from the National Intellectual Disability Database. The study consisted of a pre-interview questionnaire and a face-to-face interview. Overall the questionnaire modules included health and health conditions, healthcare utilisation, lifestyle behaviours, medications, social and community participation, occupation and activities of daily living. In their first wave of data collection TILDA included objective measurement (OM) however due to the cumbersomeness of ethics IDS-TILDA did not include OM until the second Wave. The OM process was specifically designed and under pinned by an emancipatory philosophy to ensure all participants had the opportunity to be included and empowered to engage. The OM included height, weight, grip strength, timed up and go, waist and hip circumference, blood pressure (sitting and standing) and quantitative ultrasound of the heel to examine bone health (QUS). All participants from the main study were invited to participate in the OM and to avoid connotations of being measured or tested the process was labelled '*A Health Fair*'. The data from Wave 1 was examined to establish a baseline picture of the bone health of the participants and following this, the QUS results and associated risk factors for poor bone health were examined through binary logistic regression, multivariate logistic regression and chi-squared automated interaction detector. Ethics for this study was granted by the University of Dublin Trinity College and by all the participating Service Providers. All participants provided consent and all ethical principles were applied to each stage of the research study. All data was collected and is stored as per Data Protection Act (2003).

Results

In total 753 people participated in Wave 1 which concluded in 2011, the second Wave concluded in 2014 with 708 people of all levels of ID participating. These were all invited to participate in the *Health Fair* and overall 604 people participated, with 575 people having QUS. In Wave 1, 8.1% self-reported a doctor's diagnosis of osteoporosis and 11.1% (n=43/389) had DXA (Dual energy X-ray Absorptiometry) within the previous 2 years. Over 38% were on antiepileptic drugs and a further 70% reported their physical activity

level as low. In Wave 2, similar low rates of DXA were observed however the self-reported doctor's diagnosis of osteoporosis had doubled. Of those who engaged in the QUS over 70% were identified within either osteopenic or osteoporotic range as categorised by the QUS Achilles. Overall 9 in 10 men (90.8%, n=80) and 7 in 10 women (69.4%, n=100) who presented with objective evidence of osteoporosis risk on QUS did not have a self-reported doctor's diagnosis of osteoporosis. With regards level of ID over 6 in 10 of those with severe/profound level of ID presented with QUS objective evidence of osteoporosis (62.7%, n=96). Overall 20.9% noted a prior history of fracture with the prevalence slightly higher among males than females, 21.7% (n=51) versus 20.4% (n=65) respectively. Among the strongest predictors to emerge were difficulty walking, taking antiepileptic medications, being on proton pump inhibitors and having a severe/profound level of ID.

Discussion

This study is one of the first comprehensive studies on bone health among adults with an ID in Ireland. Whilst common risk factors were seen they did not have the expected impact. Of interest in this study is that poor bone health was not exclusively confined to the older age groups and was also observed at high levels among males. Despite the fact that people with ID are known to present with higher levels of ill health, DXA screening was low. However there are no guidelines for DXA screening among people with ID and coupled with difficulties people may experience when engaging in screening it would be prudent to examine alternates, as utilised in this study, as part of an overall comprehensive bone health care package.

Men appeared at equal risk for decreased bone integrity as women in this study. Considering this and considering the potential adverse health and wellbeing poor outcomes as a results of the consequences of osteoporosis, it could be considered a missed opportunity if men are not equally considered when examining bone health as women with ID. Another strong risk to emerge was severe/profound level of ID. Those with severe/profound level of ID are possibly more likely to be non-ambulant, have higher co-morbidity and have greater dependence and health needs. It is therefore not surprising that this factor emerged however raising awareness among carer's ought to be a priority. Frequently people with severe/profound ID have communication challenges and may find it difficult to express or understand their own health need, consequently health requirements may go unmet. Other concerning issues to emerge include the specific medication use. AEDs are prevalent as a consequence of the high prevalence of epilepsy in

comparison to the general population (38% versus 1-2%). Greater awareness with regards the impact of AEDs on bone health is required with a recommendation of frequent medication reviews. This is especially prudent in light of the emergence of proton pump inhibitors (PPIs) as contributors to poor bone health and the recommendation by the American Geriatric Society to update the Beers Criteria for inappropriate medicine use among older adults, strongly recommending that older adults should not be prescribed PPIs for a period exceeding 8 weeks unless for high risk patients or specific circumstance.

Conclusion

The biomechanical structural failure which results in bone fragility is underpinned by a multitude of contributing factors. Overall a number of risk factors were identified in this study and along with the high levels of objective evidence of poor bone health the risk of fractures increases. Prevention strategies for both osteoporosis and fracture in this population should be a priority. Osteoporosis is a silent deadly disease often only realised post fracture. Despite a high prevalence of risk factors, people with ID are often less likely to be included in regular screening. By ensuring the correct supports are in place and adopting an inclusive non-threatening method of assessment or alternate method as part of an overall bone health care package such as QUS, the inevitability of bone compromise does not have to be a reality. The findings in this study supports a need for robust risk assessment and for clinical practitioners to not only consider the obvious risks but also specific concerns for people with ID in order to better target preventative strategies. Considering the potential for equal a risk of compromised bone health between women and men with ID it would be prudent to engage in further investigation specifically targeting men.

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ABBREVIATIONS

AAIDD	American Association of Intellectual and Developmental Disabilities
A&E	Accident and Emergency
ABA	Bord Altranais agus Cnáimhseachais na hÉireann (Nursing and Midwifery Board of Ireland)
ADL	Activities of Daily Living
AED	Anti-epileptic drugs
ANP	Advanced Nurse Practitioner
AOR	Adjusted Odds Ratio
ATC	Anatomical Therapeutic Chemical Classification
BAPEN	British Association for Parenteral and Enteral Nutrition
BMD	Bone Mineral Density
BMI	Body Mass Index
CAPI	Computer Assisted Program For Interviewing
CHAID	Chi-squared Automatic Interaction Detector
CI	Confidence Interval
CNS	Clinical Nurse Specialist
COPD	Chronic Obstructive Pulmonary Disease
CRF	Clinical Risk Factors
CVA	Cerebral Vascular Accident
DoH	Department of Health
DoHC	Department of Health and Children (name changed in 2011 now the DoH)
DS	Down syndrome
DXA	Dual energy X-ray Absorptiometry
ELSA	English Longitudinal Study on Ageing
FRAX	Fracture Risk Assessment Tool
GERD	Gastroesophageal Reflux Disease
HIPE	Hospital In-Patient Enquiry Scheme
HSE	Health Service Executive
ID	Intellectual Disability
IDS-TILDA	The Intellectual Disability Supplement to the Irish Longitudinal Study on Ageing
INN	International Non-Proprietary Name
IOF	International Osteoporosis Foundation
IOS	Irish Osteoporosis Society
IPAQ	International Physical Activity Questionnaire
IQ	Intelligent Quotient
MAR	Missing at random
MCAR	Missing completely at random
MUAC	Mid Upper Arm Circumference
MUST	Malnutrition Universal Screening Tool
NIDD	National Intellectual Disability Database
NMAR	Not missing at random
NOF	National Osteoporosis Foundation
NOGG	The National Osteoporosis Guideline Group
NORA	National Osteoporosis Risk Assessment
OM	Objective Measurement
OR	Odds Ratio
PA	Physical Activity
PICO	Patient/Population, Intervention, Comparison, Outcome.
PIQ	Pre Interview Questionnaire
PKU	Phenylketonuria
PPIs	Proton Pump Inhibitors
QNHS	Quarterly National Household Survey
QUS	Quantitative Ultrasound

SD	Standard Deviation
SPSS	Statistical Package for Social Sciences
TIA	Trans Ischemic Attack
TILDA	The Irish Longitudinal Study on Ageing
VIF	Variance Inflation Factor
WHO	World Health Organisation

CHAPTER 1

INTRODUCTION AND CONTEXTUALISING THE THESIS

"The ending is nearer than you think, and it is already written. All that we have left to choose is the correct moment to begin."

- Alan Moore

1.0 Introduction

This chapter introduces the reader to the thesis and begins with guiding the reader through its structure. The chapter sets the scene for the investigation of bone health among people with intellectual disability (ID). The background and justification for the study are presented and the definitions used within the thesis. The aims and objectives of the study are then introduced and the researcher maps each objective to the relevant thesis chapter.

1.1 Thesis structure and outline

Chapter 1: **Introduction and contextualising the thesis**. This chapter introduces the reader to the topic of the thesis, lays out the structure and presents the background and justification for the ensuing investigation. The chapter concludes with the presentation of the aims and objectives of this thesis and maps them to each of the study chapters. Following this the literature is presented in two chapters.

Chapter 2: **A systematic review of the prevalence of osteoporosis and osteopenia among adults with an intellectual disability**: This chapter presents a systematic investigation of the prevalence of osteoporosis among older adults with ID. The researcher presents the overall protocol developed for this investigation. This includes the database search plan, the methodical process and criteria applied along with the quality review process. From the chosen articles the researcher presents the results and summarises these in respect to the overall ID osteoporosis literature.

Chapter 3: **Contributing risk factors and challenges to maintaining strong bones among adults with an intellectual disability**: This chapter presents an exploration of the literature on risk factors contributing to osteoporosis among people with ID and explores the barriers and challenges in maintaining good health especially bone health, for people with ID.

Chapter 4: **The Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing (IDS-TILDA)**: This chapter introduces the reader to the IDS-TILDA study elucidating the sampling strategy, the underpinning conceptual and values framework that contribute to this thesis and enumerates the participation of the IDS-TILDA study. The chapter concludes with the presentation of the ethical principles that are embedded within this study that have guided and directed the researcher and this work in its totality.

Chapter 5: **An exploration of the bone health of older adults with intellectual disability in Ireland: Results from the first wave[1] of the Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing:** This chapter explores the bone status of the participants from the first wave of the IDS-TILDA study. The reported doctor's diagnosis is examined and its relationship to identified risk factors.

Chapter 6 **Osteoporosis and people with Down syndrome; a preliminary descriptive examination of the Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing Wave 1 results:** In light of the reported higher risk for osteoporosis among people with Down syndrome, the data for those who reported DS in Wave 1 was extracted and examined for reported diagnosis and an exploration of the risk factors identified.

Of note chapters 5 and 6 identified low rates of self-reported doctor's diagnosis however risk factors were evident and screening was low. The researcher concluded that further objective measurement is required to investigate bone health among older adults with ID. To that end the following chapters present, explore and investigate the objectively measured bone status of adults with ID.

Chapter 7: **Design and development of the health assessment for the Intellectual Disability Supplement to the Irish Longitudinal Study on Ageing:** With consideration of the requirement to objectively measure the bone status of this population the researcher designed a health assessment suite specifically for people with ID. This chapter presents the approach, processes and techniques employed along with the underpinning philosophy for conducting the suite of health measures which includes bone health assessment.

Chapter 8: **Exploring the objectively measured bone status of adults with intellectual disability:** Bone status was measured using a GE Lunar Achilles. Presenting the results in this chapter the investigation of the consequences of osteoporosis, namely fracture, is examined.

Chapter 9: **Predictors for poor bone quality among adults with an intellectual disability in Ireland:** This chapter presents an examination of the risk factors and predictors in relation to the objectively measured bone status.

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[1] Wave: Refers to each iteration of data collection conducted by the IDS-TILDA study, the time between waves is three years.

Chapter 10: **Bone health for life**: The final chapter provides an overview of the thesis findings and presents recommendations for clinical practice, further research, education and policy.

1.2 Background to the study

1.2.1 An ageing world

Global ageing has seen an unprecedented increase over the last number of decades. In fact, the World Health Organisation (WHO) estimates this trend to continue and by 2050 there will be 2 billion people living worldwide over the age of 60 years (Beard, 2011). In Ireland it is estimated that 21% of the Irish population will be over 65 years by 2048 (Kenny and Nolan 2014). Although most people will age well there are a significant number who will present with health challenges and it is well documented that this increase in the older population will give rise to a significant increase in chronic diseases placing a burden on the individual, the healthcare system and on society as a whole. Chronic disease includes cardiovascular disease, diabetes, cancer and musculoskeletal conditions especially osteoporosis (Department of Health and Children (DOHC), 2005). The underlying cause for many of these conditions can be attributed to lifestyle factors and other determinants of health; such as lack of exercise, excessive alcohol intake, smoking, obesity or other environmental factors. Approximately 86% of deaths and 77% of the overall disease burden in Europe has been attributed by the WHO to these chronic disease types (World Health Organization, 2002).

Ageing in poor health should not be inevitable and countries are working extremely hard at all levels in an attempt to improve the overall health and wellbeing of its citizens. Ireland has put in place a framework policy for a *Healthier Ireland* to improve the general standard of health for all its citizens (Department of Health, 2013). It is hoped that this positive initiative will contribute to the improvement in the overall health of the population especially as the ageing population increases.

1.2.2 An ageing population with intellectual disability

Until quite recently, the lifespan of people with an ID was far shorter than the lifespan of the general population. Currently there are 27,691 people with ID in receipt of services registered on the National Intellectual Disability Database (NIDD), (Kelly and

O'Donohoe, 2014). The NIDD is Ireland's national database which provides the information on service provision and requirements for planning future service need of people with ID. The data is collated in collaboration with service providers and people with ID (Health Research Board, 2013). Kelly *et al.,* (2012) note that the proportion of people with a moderate, severe or profound ID over the age of 35 years has increased from 29% in 1974 to 48% in 2012. Since 2002 the number of people living in Ireland with an ID over the age of 55 was 1,666 according to the NIDD registration records this has increased considerably to 3,675 by 2013 (Kelly and O'Donohoe, 2014). This represents a 54.6% increase in just over a decade which reflects the increasing lifespan of people with ID.

Such a remarkable success story is reflective of better education, public health improvements, economical contributions, along with medical advancements. However despite these figures the lifespan of people with ID still remains lower than the general population. Lavin *et al.,* (2006) report the average age of death in Ireland for people with ID at 46 years. Current work in the area of mortality is demonstrating that people with ID continue to die earlier (McCarron *et al.,* 2015) and mortality rates continue to be higher than those of the United States or Australia for example (Janicki *et al.,* 1999, Bittles *et al.,* 2002). It is also recognised that people with an ID are 2.5 times more likely to present with adverse health conditions than people without ID (van Schrojenstein Lantman-De *et al.,* 2000). McCarron *et al.,* (2013a) in their study report the prevalence of multimorbidity and complex conditions are greater than the general population. In their study they report that 71% of people with ID were multimorbid, defined as having 2 or more conditions, and the prevalence among the younger age group (40 – 49 years) was equal to that of the older general population (70 years and over) (McCarron *et al.,* 2013a).

Overall people with ID present with more complex and adverse health conditions, experience more health challenges (McCarron *et al.,* 2013a, Krahn *et al.,* 2006) and inequalities engaging in health care and health promotion, with needs often going unmet (Lennox and Kerr 1997, Kerr 2004, Robertson *et al.,* 2011, Martin *et al.,* 2001). One area frequently overlooked is bone health (Srikanth *et al.,* 2011).

1.3 Study rationale

Musculoskeletal disorders are the second highest cause of years lost due to disability worldwide and they account for 21% of all years lost to disability. They have the fourth greatest impact on overall health and it is estimated that worldwide 1.7 billion people suffer from musculoskeletal disorders with an estimated 45% increase likely over the next 20 years (Murray *et al.,* 2013). In contrast to the traditional health burdens of cardiovascular disease, cancer and diabetes, musculoskeletal disease is on the increase (Cooper, 2014). Part of this family of conditions is osteoporosis, however to gain an appreciation of what occurs when someone develops osteoporosis the normal pathophysiology of bone must be understood.

1.3.1 Pathophysiology of Bone

Bone is a living organ in continual flux. Bone tissue is constantly being removed and replaced and this continuous process is known as bone resorption and bone formation (bone turnover or remodelling, see *figure 1.0*) resulting in a dynamic lifetime balance of bone mass. Osteoclasts and osteoblasts are key cells in bone remodelling, osteoclasts resorb bone minerals and organic matrix and osteoblasts replenish bone matrix prior to mineralisation (Väänänen and Zhao, 2008). Bone mineral density (BMD) increases steadily from birth and reaches its peak in early adulthood, the greatest increase occurring within the pubescent years at which time the peak bone mass is reached. It is known that immature bones respond better to muscle stimuli therefore weight bearing exercise is essential in young people to promote excellent peak bone mass (Kohrt *et al.,* 1997, Walsh *et al.,* 2009). A healthy active lifestyle at this point in life is imperative due to the impact on bone health.

In adulthood around 10% of bone is remodelled each year which maintains the status of healthy bone, prevents bone fatigue damage and promotes calcium homeostasis (Seeman and Delmas, 2006). Imbalance occurs when bone resorption outpaces bone formation in many skeletal sites leading to fragility in the bone structure and a decrease in bone density. This occurs with age, especially in postmenopausal women and is known as primary osteoporosis; however secondary osteoporosis is influenced by a number of other factors (Eastell, 2009). The rate of bone turnover is affected by; the sex hormones - oestrogen and testosterone, parathyroid hormone, protein, calcium and vitamin D intake, regular weight bearing exercise and healthy lifestyle choices. Specific conditions are also

associated in manifesting poor bone mass such as, diabetes, hyperthyroidism, rheumatoid arthritis, malabsorption as a result of gastrointestinal conditions such as reflux and, epilepsy as a consequence of the required medications and their effect on bone formation (Seeman and Delmas 2006, Kanis *et al.*, 2009).

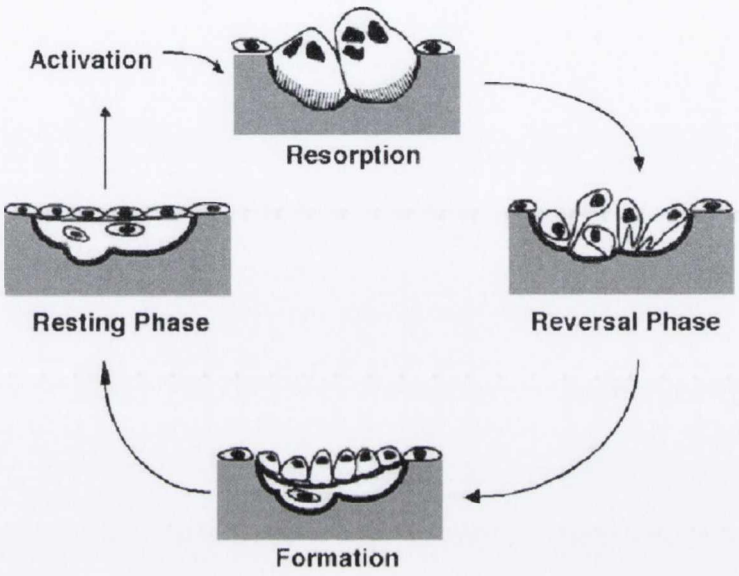


Figure 1.0 The bone remodeling cycle

Osteoclasts begin bone resorptions by removing a portion of the bone which triggers bone formation whereby osteoblasts lay down collagen and minerals over the area previously remodelled by osteoclasts, this activity maintains bone mineral density and strength.

(Idhammad and Abdali, 2012)

1.3.2 Osteoporosis and the ageing world

Osteoporosis is a chronic disease becoming more prominent on the health agenda primarily due to the debilitating consequences of this disease. Osteoporosis is insidious and initially symptom free. As such most people do not realise they have the condition until they are diagnosed perhaps post a fracture (Kanis, 2002). The deterioration of the bone

tissue leads to increased bone fragility and increases the risk of fracture particularly of the hip, vertebrae and wrist (International Osteoporosis Foundation, Compston *et al.,* 2009). The World Health Organization (WHO) (2003) estimates that currently over 75 million Americans, Europeans and Japanese, have osteoporosis. The National Osteoporosis Foundation (NOF) (2010) of the United States report that currently osteoporosis affects 9 million women over the age of 50 years with a predicted rise of one million by 2020. Consequently the annual incidence of fractures, the deleterious clinical outcome of osteoporosis, is more than 1.5 million in the US with Caucasian postmenopausal women having a greater risk of having an osteoporotic fracture in their lifetime (Lane *et al.,* 2006). Siris *et al.,* (2001) also notes in the National Osteoporosis Risk Assessment (NORA) study that even with mild to moderate low bone mass there is a significant increased risk for postmenopausal women experiencing a fracture.

In Ireland it is estimated that over 300,000 people have osteoporosis with a steady increase predicted primarily due to an ageing population (Health Service Executive (HSE) 2008). It is estimated that one in three women and one in five men may have osteoporosis (HSE, 2008). The Irish Osteoporosis Society report that osteoporosis is underdiagnosed and undertreated in Ireland with an estimated 3000 osteoporotic related hip fractures occurring annually (Kavanagh, 2015). The National Dairy Council predicted that one in four Irish women and one in 20 Irish men will suffer a fracture due to osteoporosis by the age of 60 (Condon, 2002). Subsequently the associated pain, physical disability and economic consequence suffered by people impacts on their daily living activities, quality or life and ultimately quality life years lived.

1.3.3 Osteoporosis and people with ID

Bone health underpins healthy ageing and is a major determinant of successful active ageing (World Health Organization 2003). It is reported that people with ID present with increased risk of low bone mineral density (Aspray *et al.,* 1998, Center *et al.,* 1998, Baptista *et al.,* 2005, Jaffe *et al.,* 2005). Moreover it is increasingly recognised and of urgent concern that osteoporosis continues to be under diagnosed and undertreated in the older population of people with ID (Nguyen *et al.,* 2004, Jasien *et al.,* 2012). International studies suggest that people with ID have an increased loading of risks for poor bone health (Haveman *et al.,* 2011). In particular those with Down syndrome (DS) can have poorer bone health despite this, they are less likely to have bone health checks or routine screening (Michael, 2008). Srikanth *et al.,* (2011) supports this lack of screening and specifically

highlights that the one area often neglected in physical health checks for people with ID is bone health status.

As alluded to earlier age related bone loss and osteoporosis increases the risk of fracture and subsequently can contribute to loss of independence, reduced quality of life and increased pain, morbidity and mortality for the elderly. In recent years there has been an increase in recognising these consequences and there is a plethora of bone health investigations among the general population; however this is not the case for people with ID. There is a paucity of studies on bone health among people with ID therefore the need for further investigation into this critical and growing health issue is justified.

1.3.4 Consequences of poor bone health

Osteoporosis does not in itself cause pain, it is insidious and known as the silent thief whereby bone demineralisation weakens bone asymptomatically, however this predisposes the bone to an increased risk of fracture which is the major consequence of poor bone health.

Fracture is the most common clinical outcome of poor bone health. The most common fractures are proximal femur, distal radius or vertebral fracture, leading to pain, increased morbidity and loss of independence. Eastell (2009) reports an increased risk of sustaining one of these fractures at 40% for women and 15% for men after the age of 50 years. The Irish Osteoporosis Society report hip fractures as the one fracture with the most significant consequences. The rate of hip fractures increases exponentially with age, mortality rates associated with hip fractures are approximately 20% of people over the age of 60 years predominantly due to complications such as infection, pneumonia or haematoma. Zimmerman *et al.,* (1999) reports of the high prevalence of this type of fracture among residential settings for the elderly and Gold *et al.,* (1996) describe the impact on the quality of life of people who suffer a hip fracture noting that 50% of those over 60 years will not regain their independence in their daily activities of living such as washing, dressing or walking unaided.

Considering the growing ageing population of those with ID, the clinical consequences of osteoporosis and the impact of those consequences on quality of life investigating bone health among people with ID is timely and required.

1.4 Definition of Terms

1.4.1 Defining intellectual disability

For the purposes of the study, intellectual disability is defined as a disability characterised by significant limitations of both adaptive behaviour and intellectual functioning. The internationally recognised terms mild, moderate, severe and profound ID are used as descriptive terms only. ID originates before the age of 18 years and causes limitation in many everyday functional and social skills. ID has been classified through performance on IQ tests as mild (IQ 69–55), moderate (IQ 54 - 40), severe (IQ 39 – 25) or profound (IQ <25). These cut offs are typically based on tests with a mean score of 100 and a standard deviation of 15 and do not reflect the standard error of measurement which is approximately 5 points (AAIDD, 2013).

Intellectual Disability is defined as a disability characterised by significant limitations of both adaptive behaviour and intellectual functioning



ID is a prescribed term, used within the health care profession which expresses a common understanding of what is being discussed explored or studied (Gates and Wilberforce 2003) and it is not for the purposes of labelling or describing people. Over time there have been changes in the definition of ID which reflects the transition from the more medical orientated approach to a more ecological approach whereby disability is defined in context of how the person interacts with their environment and the supports needed to enhance and maximise the person's potential. The World Health Organisation's Classification of Functioning, Disability and Health emphasis the impact of disability on the person's social context and this is reflected in the Diagnostics and Statistical Manual of Mental Disorders (DSM-V) (2013), whereby the attributed name was revised from mental retardation to intellectual developmental disability. The parenthetical 'developmental' reflecting the cognitive capacity deficits that begins in the developmental period.

1.4.2 Defining Osteoporosis

Osteoporosis describes a condition of the skeletal system. It is characterised by low bone mass and micro-architectural deterioration with a resulting increased susceptibility to fracture due to the increase in bone fragility. These fractures are most common on the vertebral body, the proximal femur and distal forearm. They are mainly associated with and commonly occur in postmenopausal women and older aged adults, this type of

osteoporosis is known as primary osteoporosis and is directly associated with age (Siris *et al.*, 2001).

Internationally the World Health Organisation's (WHO) operational definition of osteoporosis is well established and used for over 20 years. The presence of osteoporosis is defined as having a bone mineral density (BMD) value 2.5 SD or more below the young female adult mean and severe osteoporosis as a value for BMD 2.5 SD or more below the young female adult mean in the presence of 1 or more fragility fractures (Kanis *et al.*, 2008a). Originally developed to characterise the burden of disease and identify those at high risk BMD became a fundamental component of guidelines for the intervention and treatment of osteoporosis. Subsequently, BMD became the indicator for the instigation of treatment. Having BMD central to an operational definition poses problems because although there is a well-established relationship between fracture risk and low t-score, the problem that arises is that the risk is a continuous one and if the cut off of -2.5 is only considered, those with a t-score greater than this are left untreated and it is know that many fractures occur among this cohort (Kanis *et al.*, 2013). Therefore defining osteoporosis and osteopenia cannot be dependent on BMD alone but a holistic approach must be adopted when defining poor bone health.

The following present the current definition by the WHO, the definition adapted by Kanis and colleagues through their development of the fracture risk assessment tool (Kanis *et al.*, 2008b) (FRAX™) and now developed further in the National Osteoporosis Guideline Group (NOGG) programme 2012.

- WHO definition: The WHO definition is based on BMD measured by Dual-energy X-ray absorptiometry scanning (DXA) which results in a t-score. There are 3 categories, normal BMD (T-score of -0.999 and above); osteopenia (T-score of -1.0 to -2.499); osteoporosis (T-score $\leq$ -2.5) (Genant *et al.*, 1999).

- FRAX™: Is a prediction algorithm (http://www.shef.ac.uk/FRAX) that allows the estimation of a 10 year prediction of the risk of fracture. It incorporates the BMD, and 11 other clinical risk factors (CRF) of poor bone health namely; age, gender, height, weight, history of prior fracture, parental history of fracture, current smoker, alcohol $\geq$3 units/day , glucocorticoid use and rheumatoid arthritis. The purpose of its development was to inform clinical practice for the instigation of treatment for osteoporosis and inform practice that diagnosis should not be based on BMD t-score

alone, as a person with many CRF may have a higher t-score (above -2.5) but be equally or greater at risk of fracture (Kanis *et al.*, 2009, Kanis *et al.*, 2008b).

- NOGG: The National Osteoporosis Guideline Group (NOGG) is a UK based group established to provide clinical guidelines on the management of those who presented with high fracture risk. They propose to integrate the BMD measure, the FRAX output with clinical management strategies for the treatment of osteoporosis. NOGG have revised the assessment and intervention thresholds in the UK as this would increase the detection rate of individuals who could possibly fracture. They have developed a clinical management algorithm to guide clinicians for the assessment of those at risk and the implementation of intervention therapy (Compston *et al.*, 2013).

For the purposes of this study the researcher has adopted a combination of the definitions above and define osteoporosis and osteopenia as

> *A multifactorial disorder characterised by low bone mass and micro-architectural deterioration of bone tissue leading to enhanced bone fragility and a consequent increase in fracture risk.*
>
> (Kanis *et al.*, 2013, Cooper 2014, Mergler *et al.*, 2009, Srikanth *et al.*, 2011)

Osteopenia and osteoporosis are preventable but prevalent conditions among the older generation. Orthopaedic events as a result of these conditions have debilitating consequences for all older people with many experiencing reduced levels of independence and increased dependence. The economic health burden of osteopenia and osteoporosis is one of the largest health cost currently identified in the United States at an annual cost of almost $19 billion with an estimated increase in cost of almost 50% by 2025 (Burge *et al.*, 2007). Identifying the extent of the condition in people with ID would contribute to the development of prevention strategies and ameliorating interventions to reduce these preventable conditions. To that end the following study aims and objective are employed for this study.

1.5 Aims and objectives

1.5.1 Aim

The overall aim of this study is to describe the prevalence and predictors of osteopenia and osteoporosis among adults with an intellectual disability over the age of 40 years in Ireland. The study aims to identify the current status of bone health of adults with intellectual disability in Ireland, engage older adults in the objective assessment of their bone health and objectively examine the predictors of poor bone health among this cohort.

1.5.2 Objectives

The studies aims will be achieved through the following objectives. See **table 1.1** to ascertain the chapters in which these objectives are addressed:

1. To identify the prevalence of poor bone health among older adults with intellectual disability
2. To develop a baseline picture of bone health at time one of the IDS-TILDA study.
3. To develop and establish a user friendly approach to endorse the inclusion of people with intellectual disability in health assessments and particularly bone health assessment.
4. To objectively measure the bone status of older adults with ID participating in the IDS-TILDA study.
5. To examine the differences between the objectively measured bone health and a self-reported doctor's diagnosis of osteoporosis of participants in the *Health Fair.*
6. To identify modifiable and non-modifiable risk factors for poor bone health among adults with ID.

Table 1.1 Mapping the study objectives to the thesis chapters

Objective 1: To identify the prevalence and risk factors of poor bone health among older adults with intellectual disability	• Systematic review to identify the prevalence of osteoporosis among adults with ID – **Chapter 2** • Exploring the literature of the risk factors for osteopenia and osteoporosis – **Chapter 3** • Developing the baseline picture of the bone health status of adults with ID in Wave 1 – **Chapter 4** • Exploring the objectively measured bone status of adults with intellectual disability – **Chapter 8** • Examining the risk factors for poor bone health among adults with an intellectual disability – **Chapter 9**
Objective 2: To develop a baseline picture of bone health at time one of the IDS-TILDA study.	• Developing the baseline picture of the bone health status of adults with ID in Wave 1 – **Chapter 4** • Investigating the bone health of adults with Down syndrome – **Chapter 5**
Objective 3: To develop and establish a user friendly approach to endorse the inclusion of people with intellectual disability in health assessments and particularly bone health assessment.	• Designing and developing a health assessment suite suitable for people with an intellectual disability— **Chapter 7**
Objective 4: To objectively measure the bone status of older adults with ID participating in the IDS-TILDA study	• Exploring the objectively measured bone status of adults with intellectual disability – **Chapter 8**
Objective 5: To examine the differences between objectively measured bone health and doctor's diagnosis of osteoporosis.	
Objective 6: To identify modifiable and non-modifiable risk factors for poor bone health among adults with ID.	• Predictors of poor bone health among adults with an intellectual disability – **Chapter 9**

1.6 Conclusion

This chapter has briefly introduced the thesis signposting the reader to its layout and ensuing chapters. The background and justification of the study has been presented and a rationale for undertaking the study has been elucidated. The study aim and objectives have been presented and mapped to each of the thesis chapters. The following two chapters explore the literature on the prevalence and risk factors of osteoporosis among adults with ID.

CHAPTER 2

A SYSTEMATIC LITERATURE REVIEW OF THE PREVALENCE OF OSTEOPOROSIS AMONG ADULTS WITH AN INTELLECTUAL DISABILITY

"The man who removes a mountain begins by carrying away small stones."

- William Faulkner

Chapter summary

The following chapter examines the prevalence of osteoporosis among older adults with intellectual disability through the application of a systematic literature search and review. A robust protocol was developed to guide the systematic review. Predefined terms were identified and the search concepts were mapped. Guided by this map, the chosen databases which included MEDLINE, EMBASE, PubMed, PsyInfo and CINAHL were searched. The results were refined through title, abstract and finally full text screening of the remaining manuscripts. In total 50 full articles were extracted and following further scrutiny 27 went forward for quality assessment. Strict quality assessment was conducted independently by two investigators using the National Institute of Health (NIH) guidelines for assessing the literature quality. All queries were resolved by consensus and discussion between the independent investigators. Where disagreement occured a third investigator adudicated. Finally, 6 articles were chosen for data extraction. Overall the prevalence of osteoporosis was high among people with ID, especially men with ID and people with Down syndrome. However, the chosen studies predominatly targeted younger adults with more mild to moderate level of ID, and those who were mobile. Overall the sample sizes of the studies were relatively small. To conclude, further exploration is warranted, however osteopenia and osteoporosis should not be identified on bone mineral density alone, predictors and risk factors should to be examined and included.

2.0 Introduction

Peer-reviewed published literature was systematically searched and reviewed to identify the prevalence of osteopenia and osteoporosis among adults with intellectual disability. A systematic review protocol was developed to guide the search strategy, assess the quality of each of the included studies and guide data extraction. In total six articles met the inclusion criteria following quality assessment. An overall prevalence rate was difficult to establish due to the divergence in reporting method of results, just one study provided raw percentage rates of osteoporosis with two other studies providing overall t-scores for a subsample of their population under study. Coupled with this, due to the heterogeneity of samples and the variety of diagnostic instruments used to identify osteoporosis in the studies, meta-analysis was not possible. The researcher therefore applied descriptive narrative synthesis of the quantitative studies to establish the bone status of adults with intellectual disability (ID).

2.1 Background

Osteoporosis is a systematic skeletal disease that causes deterioration of the microarchitectural structure of bone leaving it susceptible to low trauma fracture (Kanis *et al.,* 1994, Kanis *et al.,* 2008a). Ireland is currently rated sixth highest in hip fracture rates in the world, singularly one of the highest causes of death worldwide (Kanis *et al.,* 2013, Hernlund *et al.,* 2013). Osteoporosis impacts on the overall health and well-being of the individual who suffers such an event. Current general population prevalence in Ireland is estimated at over 300,000 people with a steady increase predicted primarily due to an ageing population (Health Service Executive (HSE) 2008). It is estimated that one in three women and one in five men may have osteoporosis in Ireland (HSE 2008). People with an intellectual disability (ID) are living longer and subsequently are at risk of presenting with older age conditions such as osteoporosis. Identifying prevalence of osteoporosis among people with ID contributes to developing an overall picture of the status of the condition, provides information for clinical practice, policy makers and health promotion, highlights the need for focus on the particular condition and supports further investigation into the condition.

Evidence to date suggests higher rates of osteoporosis among people with ID due to co-morbid conditions such as epilepsy and use of anti-epileptic medication, mobility issues and inactivity especially for those with more severe to profound ID and lack of

exposure to sunlight reducing synthesis of vitamin D (Glick *et al.,* 2005, Guijarro *et al.,* 2008, Gonzalez-Aguero *et al.,* 2011, Dreyfus *et al.,* 2014). However a number of studies report a varied prevalence of osteoporosis, from Lin and colleagues reporting a prevalence of 26.1% (Lin *et al.,* 2015) among a cohort of institutionalised residents with intellectual disability in Taiwan, to studies such as Schmidt *et al.,* (2004) and Leslie *et al.,* (2009) reporting prevalence of osteoporosis at 67.6% and 78.5% among people with ID also living in group homes or residential settings, the former in the United States the latter in Canada. From these it is reasonable to assume that people with ID are at risk of osteopenia and osteoporosis however the reported differences vary greatly and to date there are no international consensus guidelines recommending the assessment or monitoring of bone health among adults with ID.

Investigating prevalence among people with ID will contribute to clinical practice, identify areas for further research and contribute to developing consensus on prevalence with the end goal of avoiding the clinical outcome of osteoporosis, namely fracture. The purpose of this systematic review is to establish the prevalence of osteoporosis among adults with ID.

2.2 Methods

2.2.1 Literature Review Protocol

A literature review protocol was developed to guide this systematic review (see appendix 1). The first investigator (EB) developed the protocol which includes the background, definitions, search strategy, methods and quality assessment criteria application, data extraction forms, analysis and discussion plan. In consultation with the University libriarian the first investigator consolidated the search strategy, refined the terms used and ran the initial search. The second (MOD) and subsequent investigators (JBW, PMC, MMC) reviewed the protocol and agreed with its proposed implementation. In developing the protocol the first investigator initially developed the review question using the PICO framework (Patient/Population, Intervention, Comparison, Outcome) to ensure each essential element was included (Richardson *et al.*, 1995) and is stated as:

> **What is the prevalence of osteoporosis and/or osteopenia among adults (≥18 years) with intellectual disability as defined by the AAIDD?**
>
> (Schalock *et al.*, 2010)

2.2.2 Search strategy

The databases chosen included MEDLINE, EMBASE, PubMed, PsyInfo and CINAHL using predetermined search terms for the three main concepts of the study, namely 'Bone Health', 'Intellectual Disability' and 'Prevalence' (see systematic review protocol appendix 1 for full list of terms identified and used). Full text and MeSH terms were utilised. Searches were conducted during May 2014 and search updates were created to notify the lead researcher of newly published or indexed articles meeting the inclusion criteria. The databases were searched through the three identified concepts (see *figure 2.0*). Each of the concepts relates to pre-identified terms which incorporated the initial individual searchs, these were then re-run in combination with the subject headings and key words. Finally each of the concept results were searched in combiniation with each other. Further manual searches were untaken by scrutinising the reference lists of the articles identified and including reference lists from seminal work in an effort to identify any studies not found

by the electronic serach. Last, the Cochrane central register of controlled trials was searched to identify any randomised studies that could provide data for the analysis however no randomised trials were identified.

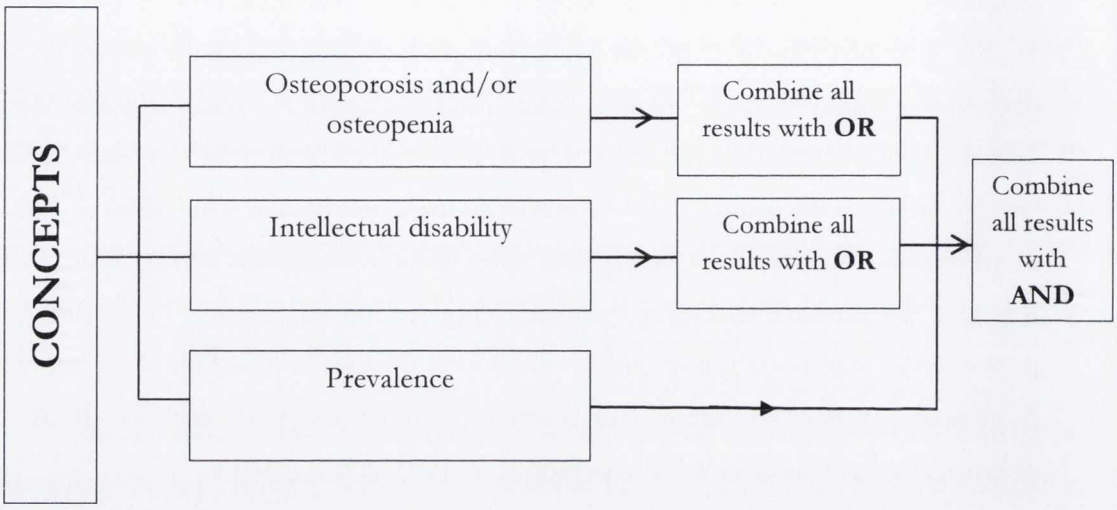


Figure 2.0 Concept search plan flowchart

2.2.3 Study selection criteria

Inclusion criteria were; published studies that were specifically pertaining to adults (≥18 years) with ID as per protocol definition, peer-reviewed publications, English language, human subjects only, no time period restrictions were placed on the search and included data on the prevalence of osteopenia and/or osteoporosis.

2.2.4 Quality assessment

The National Institute of Health (NIH) guidelines for assessing the quality of case-control studies and observational/cross-sectional studies were applied (see appendix 1:15). The NIH guidelines use 12 assessment questions for the case-control studies, and 14 questions for observational or cross-sectional studies, to assess the methodological quality of each article. Two investigators (EB and MOD) evaluated each article independently. Prior to this the investigators agreed the cut-offs prior to commencing assessment and a rating system of poor, fair and good was assigned to each of the agreed score groups. Scores of 9-12 and 10-14 were assigned for high quality studies (good), 6-8 and 7-9 for moderate quality studies (fair) and 5 or less, or 6 or less for poor quality studies (poor) (see **table 2.1**).

It was also agreed that if no high quality studies were identified studies that rated fair would be considered and a third independent investigator would adjudicate. Where any differences in item coding occurred within the quality assessment between the investigators differences were resolved through discussion and consensus. Finally, there was 100% agreement between the 2 independent reviewers regarding the articles to be included in the review following the quality scoring (**table 2.1**) (see appendix 2 for full list of articles with decisions).

Table 2.1 Quality Assessment Scores

Rating	Case-Control Studies	Observational Cohort & Cross-Sectional Studies	Action	Agreed
Good	9-12	10-14	Confirmed Yes Move to data extraction	
Fair	6-8	7-9	Adjudicate with 3rd independent investigator if no higher quality study identified and consider borderline studies (i.e. those that scored 8 for case-control or 9 for observational cohort)	
Poor	≤5	≤6	Reject	
Other	CD, NR, NA*	CD, NR, NA*	Consider adjudication where query or disagreement occurs	

*CD - Cannot determine; NR – Not applicable; NA – Not reported

2.2.5 Data extraction

Data extraction was performed using a predefined data extraction form. Rates of osteoporosis and osteopenia were recorded along with bone mineral density or bone mineral content where available for both people with ID and controls, where applicable. Additional data sought included age, gender, level of ID, aetiology, living circumstance, device used to measure bone quality and site assessed (full details can be viewed in appendix 3).

2.3 Results

2.3.1 Search results

The search and selection process is summarised in *figure 2.1* below. The initial search returned 507 papers. Duplicates were removed and the remaining identified literature were screened by title and abstract, resulting in a total of 50 articles remaining. The full text articles of each of these studies were then retieved and investigators EB and MOD applied the inclusion criteria and independently screened all full articles. Following first review 21 articles were rejected as they did not meet the selection criteria and 27 articles went forward for quality assessment. For the full list of citations those included and those excluded with justification see appendix 3. In total 6 studies were chosen as high quality studies to be included in this review.

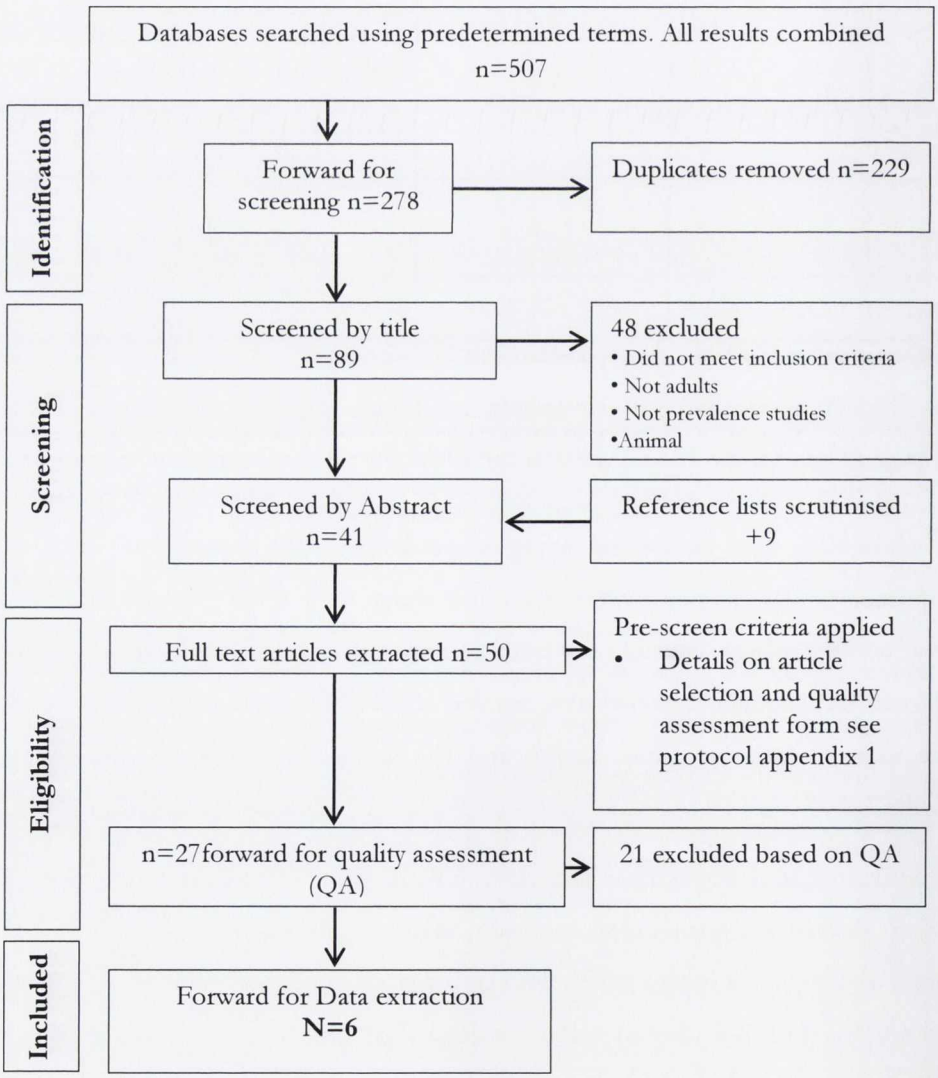


Figure 2.1 Flow diagnram for the assessment of studies identified in the systematic review

2.3.2 Summary of studies included

Of the 6 studies included in the review 2 studies were from the United States, 2 were from Greece, 1 was from Brazil and 1 from Australia. Of the studies selected 2 were published in 1998 and 1999 with a further 2 published in 2000, the remaining were published in 2010 and 2014 respectively. The study design included four case-control studies and two observational cohort studies. Two of the studies were limited to males only with the remaining four studies including both males and females. With regard level of ID, two of the studies reported on those with mild/moderate level of ID only, three studies did not specify level of ID and the remaining study noted that those with mild/moderate and severe level of ID were included, noting that none of the subjects included in their study had a profound level of ID. The average age of the participants with ID included in all the studies was 38.76 years[2] with an overall number of participants ranging from 16 to 107 people with ID participating in all the studies. Three of the studies had samples less than 50, Angelopoulou *et al.,* 1999, Angelopoulou *et al.,* 2000 and da Silva *et al.,* 2010, (N= 22, 16 and 30) respectively, two of the studies had samples below 100, Geijer *et al.,* 2014 and Center *et al.,* 1998 (N= 66 and 94) with the final study, Tyler *et al.,* 2000, reporting a sample of N=107 respectively. One of the studies examined those with Down syndrome (DS) specifically, with a further 4 studies noting that a percentage of their sample included people with DS, the remaining study did not specify any particular aetiology. All except one study (Tyler *et al.,* 2000) reported that their sample were independently mobile with 3 studies (da Silva *et al.,* 2010, Geijer *et al.,* 2014, Center *et al.,* 1998) noting this as a requirement. A description of the 6 studies included in this review can be viewed in **table 2.2**)

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[2] Overall average age was determined as follows: Sum (mean age multiple by sample no.)/total sample

$$\frac{12983.9}{335} = 38.76 \text{ years (weighted mean)}$$

Table 2.2 Description of the 6 studies included in the systematic review

Study	Design	Participants		Gender M/F		Age Mean (SD)		Level of ID	Aetiology	Independent ambulation
		People with ID (n)	Controls (n)	People with ID	Controls	People with ID	Controls			Y/N
Angelopoulou *et al.*, 1999	CC	22	27	9/13	12/15	24.93 (±3.84)	25.93 (±3.19)	Not specified [NS]	DS only	Y
Angelopoulou *et al.*, 2000	CC	16	10	16/0	10/0	23.69(±3.9)	25.0(±3.3)	Mild/Moderate	DS & other ID	Y
Center *et al.*, 1998	OC	94	-	41/53	-	35(±8)	-	Mild/Mod/Severe	DS & other ID	Y
da Silva *et al.*, 2010	CC	30	15	30/0	15/0	28.44(±6.66)	24.57 (±3.87)	Mild/Moderate	DS & other ID	Y
Geijer *et al.*, 2014	CC	66	33	30/36	15/18	43.7(±5.5)/44.2(±6.0)	43.7(±5.9)	NS	DS & other ID	Y
Tyler *et al.*, 2000	OC	107	-	61/46	-	47(±5.6)	-	NS	NS	N

CC=case control
OC=observational cohort
NS=Not specified
DS=Down syndrome
ID=Intellectual disability

2.3.3 Prevalence of osteoporosis and osteopenia

With the evolution of bone densitometry differences in technologies, acquisition techniques, reference databases, reporting methods, and terminology, have developed. As can be seen in this review no two studies used the same device (dual energy X-ray absorptiometry-DXA) to obtain the reported BMD, one study reported the BMD results in g/cm3 (Geijer *et al.,* 2014) with a further 2 studies reporting t-scores for subsamples of their study participants along with the BMD results of the remaining participants (Angelopoulou *et al.,* 2000, Angelopoulou *et al.,* 1999). With the diversity in BMD reporting and the variety of devices used, the reference cut-offs determining osteoporosis could possibly differ especially between the various devices, subsequently making it difficult to undertake direct comparison across studies or pooling data. Therefore for the purposes of this study a qualitative report of the results will be presented. Full description of the results can be viewed in **table 2.3**.

Overall, people with ID were identified as having lower BMD than their non-ID counterparts except for Angelopoulou *et al.,* (2000) who reported no significant difference between those with ID (and without DS) in comparison with the controls. However for those with DS in this study BMD was significantly lower than for non-DS individuals (*p*-value=0.002). Angeloupoulou and colleagues (2000) reported those with a DS as having a t-score of -2.66 ($\pm$0.40) which places this group of individuals with DS within the osteoporotic range. All the studies that reported results for those with DS identified a statistically significant association with low bone density.

For Angelopoulou *et al.,* (1999)'s study, BMD was significantly lower than the control group (p-value<0.0001) for males with DS, with a percentage decrease of 25% versus the control males, their mean t-score is reported at -2.62($\pm$0.82) and whilst Angelopoulou and colleagues (1999) reported females with DS as having BMD significantly lower than controls (p-value<0.001) their percentage difference was higher than the males at 14% with a mean t-score= -1.56($\pm$1.02) placing the females within the osteopenic range of bone quality. The findings from Geijer *et al.,* (2014) support these findings insofar as those with DS in their study have significantly lower BMD of the spine, hip and total body when compared with their non-ID controls, however they report those with ID (non-DS) did not have significant differences in BMD of the spine in comparison to those without ID. This differs from Angelopoulou *et al.,* (1999) and Angelopoulou *et al.,*

(2000), however da Silva and colleagues found no differences between those with ID and their control group (da Silva *et al.,* 2010). People with DS were reported separately to those with ID and were identified as having significantly lower BMD than those with ID and the control group without ID (p-value<0.001) (da Silva *et al.,* 2010), however the authors make no distinction between genders. On the other hand Center *et al.,* (1998) identify DS as an independent predictor for low BMD in women. They report their final BMD as z-score, which is the number of standard deviations above or below the BMD score normally expected for someone of the same age, gender and ethnic or racial background. However it must be noted that in the Center *et al.,* (1998) study the sample size for those with DS is n=12 females and n=8 males with DS. Similarly the final study included in this review, Tyler *et al.,* (2000), reports a low percentage of people with DS in their study. Of the total number of participants (N=107), 15% reported DS (n=16). Tyler *et al.,* (2000) is the only study to report an overall prevalence using the World Health Organisation designations of normal BMD. They found 34% with a t-score <-2.5 placing this cohort in the osteoporotic category, 21% was osteopenic and the remaining 45% registered within the normal category. Interestingly this study specifically reports that those individuals who were independently mobile had significantly higher BMD scores by comparison all the other studies excluded those who were unable to mobilise independently.

Table 2.3 Prevalence of osteoporosis and osteopenia among people with ID and controls

Study			People with Intellectual Disability				Controls			
	DXA details	Sites	Osteoporosis *n* (%)	Osteopenia *n* (%)	BMD (SD) g/cm²		Osteoporosis *n* (%)	Osteopenia *n* (%)	BMD (SD) g/cm²	
					Male	Female			Male	Female
Angelopoulou *et al.*, 1999	Lunar DPX v3.6	LV: L2-L4	mean t-score -2.62 (±0.82) Males DS	mean t-score -1.56(±1.02) Females DS	0.931(±0.114)	1.027(±0.132)	-	-	1.243(±0.072)	1.197(±0.081)
Angelopoulou *et al.*, 2000	Lunar DPX	LV: L2-L4	mean t-score -2.66 (DS only)	-	0.926(±0.07) (DS only) 1.228(±0.14) (other ID)	-	-	-	1.239 (±0.06)	-
Center *et al.*, 1998	Norland XR-26	LV: L2-L4	-	-	1.01(±0.17) [DS] 0.88 (±0.16) z-score -0.16	0.97(±0.19) [DS]0.87 (±0.12) z-score -0.44	-	-	-	-
da Silva *et al.*, 2010	DPX-IQ v4.6A	LV: L2-L4	-	-	0.94(±0.11) (DS only) 1.26(±0.14) (other ID)	-	-	-	1.28 (±0.09)	-
*Geijer *et al.*, 2014	Does not specify	Whole body	-	-	1.10 (±0.11) (DS only)** 1.20 (±0.14) (other ID)**		-	-	1.28 (±0.14)**	
Tyler *et al.*, 2000	Lunar PIXI	Calcaneus	22 (21%)	36 (34%)	0.42 (±0.12)	0.48 (±0.14)	-	-	-	-

BMD=Bone mineral density
DXA=dual energy X-ray absorptiometry
LV=Lumbar vertebrae

**Reported BMD in cm³ - totals reported here*
***Results for both M/F combined*

2.4 Discussion

All six studies of this review report that the BMD of people with ID is lower than the general population, however Geijer *et al.,* (2014) noted that they did not find a significant difference in BMD of the spine compared to those without ID, they do point out that they undertook measures of the total spine as opposed to the lumbar spine only which may account for the difference. Similarly, Angelopoulou *et al.,* (1999) report no statistical significant difference in BMD of men without DS in their study.

Nevertheless the evidence to emerge from this review demonstrates there is an alarmingly high percentage of people with ID with compromised bone integrity, increasing their risk of fracture (Kanis *et al.,* 2009a). The impact of fracture can have devasting consequences on quality of life and increase disability-adjusted life years (DALYs). Murray *et al.,* (2013) highlight the rising global burden on life years from musculoskeletal illhealth. For people with ID, who may already be experiencing physical impairment, the impact of a fracture may further limit their independence subsequently increasing their dependence and reliance on others. Recovery from fracture is challenging however for people with ID it can be even more difficult due to their underlying coginitive impairment. Other issues that can interfer with their recovery include poor compliance with physiothearpy regimes and difficulty in adhereing to health promotion due to limited or lack of education (McCarron *et al.,* 2011). These factors culminate in increasing the vulnerability of people with ID and emphasising the importance and need for further investigation of the bone health of people with ID.

Very often osteoporosis is only considered as a condition of old age with many screening recommendations being instigated after, for example age 50 years for post menopausal women or after 70 years for men where no contraindication exsist (Irish Osteoporosis Society, 2012). The people in the included studies were young and relatively active individuals with a number of studies highlighting their participant involvement in exercise programmes, groups who would not normally be associated with bone compromise making the study indications of overall low BMD a concern. Therefore considering the age groups in the studies were young (mean age 38.76 years) it is possible to assume that poor bone quality among people with ID is not necessarily a condition of old age however likely to increase in prevalence with ageing.

Another finding to emerge is the high levels of low bone density among men. Mistakeningly osteoporosis can sometimes be exclusively associated with women and may not be considered a condition that would effect men, especially those of a young age group. However the findings here indicate that men are likely to be at high risk of osteoporosis. Therefore, when planning overall health care among people with ID osteoporosis must be considered a risk for both women and men of all ages. However it must be remembered that the sample sizes of the included studies in this review overall were small and further investigation is warranted.

Osteoporosis among people with ID is no necessarily a condition of old age however is likely to increase in prevalence with advancing age



Another concern to emerge is that the investigations for low BMD are largely among adults who were mobile and among those with mild or moderate level of ID. This means that those with more severe/profound ID and those with mobility issues are omitted. It is known that those with more severe/profound ID are more likely to present with multmorbidity (McCarron *et al.,* 2013a), polypharmacy (O'Dwyer *et al.,* 2013) and immobility (Emerson, 2005), all contributing factors that compromise skeletal integrity. These omissions greatly increase the possiblity that the levels of low BMD overall among people with ID are underestimated.

All studies illustrated that individuals with DS had significantly lower BMD than people with ID or control groups without ID. Down syndrome emerges as an independent predictor of poor bone health. This is not surprising considering that people with DS very frequently present with hypogonadism, high levels of thyroid disease, immaturity in bone formation as they progress from adolescence to adulthoood, a feature of DS, and frequently people with DS are less active than their peers, all culminating in increasing their susceptibility to bone compromise and poor bone integrity (Seeman, 1999, Prasher, 1999, Hayes and Batshaw, 1993). Down syndrome appears to carry unique features that increase suscepitiblity to osteoporosis a factor that requires further investigation.

2.5 Limitations

This review set out to establish the prevalence of osteoporosis among adults with ID however due to the variety of devices used to establish and measure the BMD in the chosen studies determining an overall prevalence was not possible. Therefore the aim of this paper was not achieved however this review did establish that low BMD is a feature

among people with ID and requires further investigation, and that those with ID especially those with DS are particularly susceptable to low BMD. The findings of this review must be viewed in light of the sample sizes of each of the studies identified. Overall the samples in these studies were relatively small indicating that generalisability is not possible. However that said, the review appears to raise more questions than it answered. Questions such as; Why is BMD generally low among people with ID? What are the driving factors for low BMD? How do these factors contribute to the development of osteopenia and osteoporosis? What is the prevalence of osteoporosis among people with ID? Further investigation is warranted to address these questions.

2.6 Conclusion

Poor bone health is a concern among people with ID and ought to be investigated further, on larger samples, across all levels of ID especially those with severe/profound ID who would possibly be most at risk due to higher levels of immobility and co-morbidity. From these studies overall prevalence is difficult to establish, nevertheless it is evident that bone quality among people with ID is compromised. Considering the levels of low BMD identified among the younger cohort it is of great concern and requires further investigation to lead to action. Identifying and examining osteopenia and osteoporosis without due consideration to predictors and risk factors is impractical, therefore the following chapter explores the literature on the determining risk factors for osteopenia and osteoporosis.

CHAPTER 3

CONTRIBUTING RISK FACTORS AND CHALLENGES TO MAINTAINING STRONG BONES AMONG ADULTS WITH INTELLECTUAL DISABILITY

"Take pride in how far you've come. Have faith in how far you can go. But don't forget to enjoy the journey."

- Michael Josephson

Chapter summary

This chapter examines the literature pertaining to risk factors for osteopenia and osteoporosis among adults with ID. Subdividing the risks into modifiable and non-modifiable the researcher describes the increased burden of risk factors experienced by people with ID. The life expectancy of people with ID has extended and as such older aged health conditions, such as osteoporosis, are becoming more prevalent. Factors that directly and indirectly impact on bone health are frequently identified from syndrome specific effect such as those with Down syndrome, to health conditions such as epilepsy that increase the risk of falls as well as the required medication impacting on bone metabolism. There are also a number of modifiable factors observed among people with ID such as poor lifestyle choices resulting in obesity and inactivity to poor dietary choices contributing to potential vitamin and mineral imbalance. Other factors explored include barriers to accessing adequate health care, from communication difficulties to inadequate environmental adjustment to accommodate people's needs. In conclusion the researcher notes that although some risk factors are unalterable those factors that are amenable to change require attention to ensure a positive and healthy outcome for all those with ID.

3.0 Introduction

It is reported that for people with intellectual disability there is an increased loading of risk that predispose them to developing osteopenia and osteoporosis. These increased risks are reported to be outside the common accepted risk factors, such as smoking, increased alcohol consumption, parental history and specific corticosteroid use, as seen for the general population. With a growing older population of people with ID increased prevalence of chronic conditions will be identified. For people with ID areas such as polypharmacy and poorer lifestyle choices put this population at increased risk of suboptimal bone health. In addition people with ID face additional challenges in maintaining their own health and accessing healthcare screening. This chapter explore the risks through the non-modifiable and modifiable identified risks and further literature is expounded in subsequent chapters.

3.1 Identifying and categorising predictors of osteopenia and osteoporosis

Osteoporosis is a significant health and socioeconomic threat to a large proportion of the population. As previously noted it is a chronic condition characterised by low bone density and microarchitectural deterioration of bone tissue increasing the risk of fracture especially among older adults (Åkesson *et al.,* 1993, Kanis *et al.,* 2009, Kanis 1994, Cooper *et al.,* 2011). In recent years there has been increased attention on osteoporosis and its potential negative consequences for older people. New assessment tools, national guidelines, health promotion awareness campaigns and assessment of lifestyle factors as indicators of at risk groups have been introduced (Irish Osteoporosis Society 2012, Guessous *et al.,* 2008, World Health Organization 2003, Health Service Executive 2008). However, the range of studies conducted has predominantly been undertaken within the general population.

A literature search using CINHAL, PubMed, EMBASE and psychInfo, identified that there is a paucity of available literature pertaining to bone health and ID in comparison to the plethora of general population literature and large scale investigation studies. This exploration of the literature pertains to risk factors associated with osteopenia, osteoporosis and people with ID as well as the factors that increase their overall health burden. Following extensive searches through the databases noted above, the WHO and International Osteoporosis Foundation website a broad number of risk factors were identified, grouped and classified into two groups as follows

1. Non-Modifiable Risk Factors
    - Direct non-modifiable factors
    - Indirect non-modifiable factors

2. Modifiable Risk Factors
    - Lifestyle behaviours
    - Barriers to healthcare
    - Inadequate built environment
    - Impairing attitudes

3.2. Non modifiable risk factors

3.2.1 Direct non-modifiable risk factors

These include female gender, increased age, family history of osteoporosis, Caucasian and history of prior fracture. Late menarche, early menopause, and low endogenous oestrogen levels are also associated with low bone density (Kanis 2002) which contributes to fragility and increases the risk of fracture.

Osteoporosis across the life span

Greater life expectancy is now an established fact among the population of people with ID and with this older growing generation is an associated increased risk of osteoporosis (Center *et al.,* 1998, Petrone 2012). Peak bone mass is attained in early adulthood and remains constant into the mid to late 30's after which bone mineral density (BMD) begins to decline, thus low bone density is associated with ageing. International studies provide clues that individuals with ID have a greater variety of health care needs compared to those of the same age and gender in the general population (Haveman *et al.,* 2011). It is also important to acknowledge a common misperception that osteoporosis is always the result of accelerated bone loss. Bone loss commonly occurs with ageing however, an individual who does not reach optimal bone mass during childhood and adolescence may develop osteoporosis without the occurrence of accelerated bone loss. People with Down syndrome (DS) are known to be within this category due to underdeveloped bones of the upper spine and neck (Hayes and Batshaw 1993). Hence bone growth below the optimal levels in childhood and adolescence is just as important a contributory factor to the development of osteoporosis, placing this population at unique risk as they age.

Osteoporosis and gender

It is well documented that osteoporosis occurs in both genders (Anderson *et al.,* 1998, Barrett-Connor *et al.,* 2005) however women are at higher risk of osteoporosis simply because their bones are smaller. It is increasingly recognised that oestrogen inhibits periosteal apposition and as a result their bones are narrower than men (Seeman 2002). Additionally females are more susceptible to related fractures due to such factors as less muscle mass, fluctuation of oestrogen levels and menopause, all having an enormous impact on bone density (Melton 2003).

Women can experience a rapid decline in BMD after the menopause because of a drop in oestrogen levels and females with ID are no different (Arvio *et al.,* 2009). Specific syndromes have been associated with late menarche and early menopause both of which contribute to lower oestrogen level and hence lower BMD. Knickmeyer *et al.,* (2006) investigated a group of women with autistic spectrum disorder and, although the study sample was relatively small (N=38) and included only Caucasian females, they did conclude that menarche was delayed in these females by an average of 8 months compared to their control group counterparts. Females with DS are also known to presents with late menarche and early menopause resulting in oestrogen deficiency and consequently adverse skeletal health (Carr and Hollins 1995). In their examination of the menstrual patterns of 45 females with DS, Carr and Hollins (1995) note that 87% presented with menopause by the age of 47 years and all female participants in their study by the age of 51. This is further supported by Schupf *et al.,* (1997) who notes that the menopausal onset occurs at a much earlier age for females with DS than generally noted among females. Rödström *et al.,* (2003), for example, notes the mean age of onset of menopause for Caucasian women to be between 50 – 52 years however it can occur earlier or later. It must also be noted that oestrogen and androgens play an important role in the bone maturation of men (Krassas and Papadopoulou 2001). Hypogonadism is associated with reduction in pubertal growth spurt thus low oestrogen and testosterone levels which are prevalent among males with DS (Seeman *et al.,* 1999, Hsiang 1987) which very likely contributes to compromising the bone health in men with DS.

Osteoporosis and health complexities

Secondary health conditions and complex mobility needs are prevalent among people with severe or profound ID (Cooper *et al.,* 2007, Emerson *et al.,* 2000). These compromises

occurring early in life increase the likelihood of poor bone health. It must be noted also that presentation of osteoporotic fracture, especially those of the vertebrae, may not be clinically evident among this cohort as people with ID especially among the severe and profound level may not have verbal ability to complain of pain, subsequently fracture occurrence can be overlooked and untreated. Level of ID therefore can have adverse effect on bone health. It has also been noted that people with more severe to profound ID are more likely to present with increased co-morbidities (McCarron *et al.,* 2013b), greater complex need and, are more likely to experience fracture partly due to the higher incidence of epilepsy and falls (Glick *et al.,* 2005). *The Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing* (IDS-TILDA) identified an epilepsy prevalence rate of over 31% among older adults with ID in Ireland (McCarron *et al.,* 2011). Due to seizure activity the likelihood of falls and subsequent fracture is increased. However, it is the associated antiepileptic medication (AEDs) usage that is most damaging to bone. The enzyme-inducing AEDs include carbamazepine or phenytoin which increases the vitamin D catabolism reducing its availability to aid calcium absorption. Further, a side effect to the non-enzyme inducing AEDs, lamotrigine or topiramate for example, interfere with intestinal calcium absorption and cause calcitonin deficiency and resistance to parathyroid hormone (Babayigit *et al.,* 2006) culminating in adverse effect on bone.

Osteoporosis and syndrome specific risks

Also among the non-modifiable risks are syndrome specific risks associated with ID. For example, people with DS are particularly at risk of poor bone health and indeed Angelopoulou *et al.,* (2000) has cited DS as a contributory risk factor alone. Hypotonicity is characteristic of this syndrome increasing their risk to long bone fracture, hip dislocation, compression fracture of the vertebral body and atlantooccpital instability (Hayes and Batshaw 1993, Van Allen *et al.,* 1999, Srikanth *et al.,* 2011). Thyroid hormone (T3) is required for normal bone development in childhood and regulates bone remodelling as an adult. People with DS have higher prevalence of endocrine disorders such as hypothyroidism (Jaffe *et al.,* 2005). Karlsson *et al.,* (1998) in their longitudinal study identified a 33% development of hypothyroidism before the age of 25 years. The significance of this is that overproduction or underproduction of thyroid hormone has a profound effect on bone development and subsequently on bone health, increasing the possibility of reduced BMD. As people with DS age the prevalence of Alzheimer dementia increases rapidly (McCarron *et al.,* 2013b), the associated epilepsy, increased risk to wander and fall, medications and increased difficulty swallowing combined with the already

underlying risk of phenotype, predispose this cohort to a high risk of developing poor bone health.

Another example of syndrome specific risk are those with cerebral palsy (CP) and ID (Mergler *et al.,* 2009). Early childhood is a critical time for skeletal growth and bone mass accrual. Children with CP present with oral-motor dysfunction which effects swallowing leading to feeding difficulties and poor nutrition. Poor or absent mobility is notable, recurrent pulmonary infections and gastro-oesophageal reflux all frequently recognised among those with CP can interfere with calcium and vitamin absorption. In turn this interferes with the attainment of maximum bone density, predisposing people with CP and ID to increased risk of osteoporosis and subsequent fracture (Henderson *et al.,* 2002, Stevenson *et al.,* 2006). Prader-Willis Syndrome is another example of genetic dysfunction predisposing the person to poor bone health. People with Prader-Willis syndrome present with abnormal growth, over eating disorders resulting in obesity and overall poor and imbalance diet, hypotonia and endocrine disorders are common, culminating in the compromise of their skeletal integrity and reducing bone mineral density (Sinnema *et al.,* 2011, Burman *et al.,* 2001). *Figure 3.1* summaries in graphical form the overall non-modifiable risks for poor bone quality associated with adults with ID.

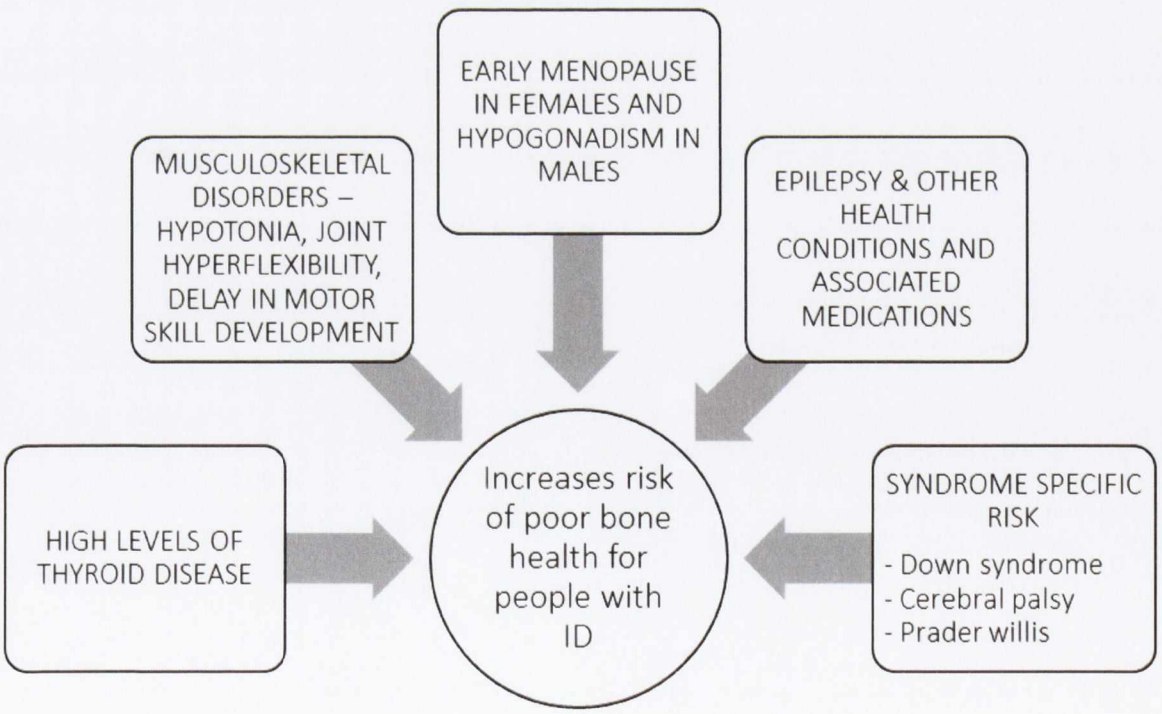


Figure 3.0 Presentation of the increased burden of risk factors for people with ID

3.2.2 Indirect non-modifiable risk factors

International studies suggest that people with ID are at greater risk of experiencing overall poor health with conditions such as epilepsy, mental health conditions gastro intestinal disease and osteoporosis to name but a few as well as higher levels of medication use and polypharmacy identified at greater rates than the general population (O'Dwyer *et al.,* 2013, McCarron *et al.,* 2011, Haveman *et al.,* 2011, Ouellette-Kuntz 2005). Prevalence rates for epilepsy among adults in the general population differ greatly from rates among those with ID, for example Linehan *et al.,* (2010) report rates of 10 per 1000 of the population over the age of 18 years in Ireland compared to 36% in those over 40 years with ID (almost 4 in 10) reported by McCarron *et al.,* (2014). Certain high prevalence can be linked to specific syndromes for example Alzheimer's dementia and Down syndrome (Tyrrell *et al.,* 2001). Mental health prevalence is reported at greater levels among those with ID in comparison to the general population, for example with regards those self-reporting doctor's diagnosis of depression 18.5% (McCarron *et al.,* 2011) versus 5% (O'Regan *et al.,* 2011) respectively. With regard dental health, Mac Giolla Phadraig *et al.,* (2014) note that despite regular contact with their dentist people with ID tend to have less preventative or restorative care. Sensory impairment can go unnoticed in people with ID especially for those with severe or profound ID with some studies reporting prevalence rates of over 50% (Evenhuis *et al.,* 2001). In that study undiagnosed cases, 90 visual impairment new cases (14.7% of 611) and 128 auditory impairment new cases (22.5% of 567) were identified, highlighting the difficulties in auditory and ophthalmological assessment among people with ID. McCarron *et al.,* (2014) also report high prevalence of sensory conditions for example cataracts substantially increased from Wave 1[2011] to Wave 2 [2014] of the IDS-TILDA study (13% to 19% respectively). Although visual impairment is associated with ageing the rates identified by TILDA (Cronin *et al.,* 2011) among the general population were somewhat lower at 11%. The challenge with visual impairment is that is directly correlated to falls and subsequent fractures, identifying such impairment as part of the overall health promotion care package is imperative.

It can be seen that overall, people with ID have higher levels of disease, can present with multi-morbidity and their health needs are greater (Cooper *et al.,* 2004, Melville *et al.,* 2006, McCarron *et al.,* 2013b). Uncommon patterns of multi-morbidity frequently occur (McCarron *et al.,* 2013a) leading to conditions being overlooked and in turn increasing the vulnerability of the person with ID which can lead to needs going

unmet and can result in premature death (Nocon *et al.,* 2008, Heslop *et al.,* 2007). Preventable premature mortality among people with ID has been highlighted in reports such as '*Confidential Inquiry into Premature Deaths of People with Learning Disabilities*' (Heslop *et al.,* 2014) and it was noted that cause of death in many cases was most likely preventable despite the overwhelming evidence of increased health need.

3.3 Modifiable

3.3.1 Lifestyle behaviour

Several modifiable behavioural factors contribute to the development of poor bone health namely, sedentary lifestyle or inactivity resulting in overweight or obesity, poor dietary habits resulting in an imbalanced diet, reduced sunlight exposure, alcohol intake of 3 units or more per day and smoking.

Physical activity is essential to maintaining health and reducing the risk of chronic disease. Physical activity stimulates bone turnover and reduces risk of poor bone health, increases muscle mass and strength thus reducing the likelihood of falling. For people with ID low levels of physical activity and a sedentary lifestyle are frequently common, are at levels that do not yield health benefit and are at lower rates than the general population. In fact people with ID engage less in physical exercise and have lower social outlets than their non-ID counterparts (Robertson *et al.,* 2000, McCausland *et al.,* 2015). Robertson *et al.,* (2000) identified an 80% rate of inactivity according to the Health Survey for England criteria. Likewise Beange *et al.,* (1999) identified similar inactivity levels as did McCarron *et al.,* (2011) in the first Wave [2011] of their longitudinal study. In the second wave [2014], the IDS-TILDA study note the trends of low physical activity levels continue, and there are marked differences in levels of physical activity between the general older population figures reported by TILDA, and the participants of the IDS-TILDA study (McCarron *et al.,* 2014). Over 73% of those with ID engaged in low levels in comparison to just over 32% of those in the TILDA study (Nolan *et al.,* 2014).

Throughout the world obesity has become a significant concern and challenge for health care providers and policy makers alike. Unhealthy lifestyle choices, especially with regards diet and exercise, impacts directly on weight and weight management leading to subsequent overweight and obesity. Currently Ireland as a nation is one of the most obese in Europe (World Health Organisation 2015a). Unfortunately people with ID are not excluded from this epidemic with significant evidence that obesity levels are highly

prevalent among the population of people with ID and particularly, women with ID and people with DS (Yamaki, 2005, Rimmer *et al.,* 2010). In the second wave of the IDS-TILDA study [2014], objective evidence points to more than two thirds of the cohort falling within the overweight and obese categories. What is of concern it that those who were more likely to be within the obese category tended to be females, within the younger age group (40-49 years) and with a more mild level of ID (McCarron *et al.,* 2014). Perhaps it is poor food choices of what to eat that contributes to this high risk (Adolfsson *et al.,* 2008), or a lack of nutritional education, or an inability to cook or prepare their own meals, it may simply be the fact that people with ID have never gotten the opportunity to learn and develop the skills of good dietary habit that contribute to these high levels of obesity. Nevertheless one or a number of these factors do contribute to eventual poor food and lifestyle choices (Melville *et al.,* 2007, Robertson *et al.,* 2000). Consequently the deviation from a healthy weight results in insufficient or imbalanced dietary intake of vitamins and minerals which impacts greatly on the status of bone health.

Living in restricted environments appears to contribute to the problem. Emerson (2005) noted that overall people with ID living in supported accommodation were at an increased risk to being underweight and inactive however found that women with ID were more at risk of being overweight to obese. Those who present as underweight similarly will have inefficient dietary intake to maintain healthy bones and preserve overall good health. Frequently people with cerebral palsy (CP) and ID present with eating and swallowing difficulties increasing their potential to be underweight along with the complex nature of CP exposing this cohort of people to poor bone health, reduced skeletal integrity and increased susceptibility to fracture (Mergler, 2012). Overall these poor dietary choices along with reduced physical activity (Emerson, 2005) predispose people with ID to an unhealthy lifestyle, illness and the development of chronic disease. The overall increase of risk of poor bone health is evident considering obesity levels which is an outcome of inactivity and unfortunately prolonged inactivity is prevalent among people with ID (Taylor and Hodapp, 2012, Beange *et al.,* 1999). By advocating for greater involvement of direct care providers as part of an amelioration plan obesity and its consequences can be addressed (Rimmer, 2011, Rimmer and Yamaki, 2006).

Apart from the implication of an imbalanced diet, obesity has been indicated in a decreased bioavailability of vitamin D (Wortsman *et al.,* 2000). In their study of cutaneous vitamin D3 synthesis, they concluded that the subcutaneous fat sequestered more vitamin

D3 and this occurred in higher rates among people who were obese, possibly because there was more adipose tissue available. Vitamin D is essential for bone metabolism as it promotes calcium absorption. Another compounding factor for those living in the northern hemisphere is the fact that sunlight hours are much shorter, reducing the potential to synthesising vitamin D from UV exposure (Holick 2003). More than 90% of most people's requirement for vitamin D is acquired from sunlight, however given the increased risk of skin cancer associated with sun exposure people are reluctant to engage in sunbathing without sunscreen. Foods such as oily fish contain natural vitamin D however Ireland overall does not have a good reputation for eating oily fish, possibly supplementation is the answer. In fact in Ireland the Food Safety Authority has recommended supplementation for all citizens (Food Safety Authority 2011). A Cochrane review recommended combining vitamin D with calcium supplementation as vitamin D alone showed inadequate effect on improved outcomes and prevention of fracture (Avenell *et al.,* 2009, Avenell *et al.,* 2014). Another factor to impact on vitamin D synthesis is institutional living. This type of living circumstance has been correlated to reduced sunlight exposure and subsequently an increase in vitamin D deficiency (Hirani and Primatesta 2005). Consequently, people with ID living in residential type settings and larger community group homes are quite possibly at risk of deficiency.

3.3.2 Barriers impacting on healthcare

For many decades now health and healthcare inequalities among people with ID have been highlighted (Krahn *et al.,* 2006, Alborz *et al.,* 2005, Kerr 2004). Factors that contribute to inequality include healthcare practitioners being poorly informed and lacking knowledge about ID, inappropriateness or lack of available health care information for people with ID, fragmentation and discontinuity of service provision especially for those with multiple and complex needs, and lack of empirical evidence on health determinants to underpin healthcare policy (Tracy and McDonald, 2015, Emerson and Hatton, 2013, Alborz *et al.,* 2005, Sowney and Barr, 2004). These factors are further exacerbated by poor physical access to services, lack of reasonable adjustment by healthcare providers, behaviours that challenge and communication difficulties of expression and interpretation experienced by people with ID (Turner and Robinson 2011, Martin 2003, Felce *et al.,* 2008, Alborz *et al.,* 2005, Hunt *et al.,* 2001, Iacono and Sutherland 2006). Communication difficulties and literacy challenges limit the capacity for the person with ID to relay their health concerns and needs to others and exclude them from mainstream health promotion. People's

cognitive skills to self-report are very often curtailed impacting on the conversational interaction between the clinician and patient. McCarron *et al.,* (2011) noted that one area people with ID report having the greatest communication challenges is with healthcare professionals. Subsequently people with ID are dependent on healthcare by proxy.

3.3.3 An inadequate built environment

Many unrecognised barriers in architectural design impede people with ID. Often assumptions are made regards literacy and people's capability to read signs, open doors, reach buttons or see obstacles (Berkman *et al.,* 2011). These disparities impinge on the person's self-efficacy (Kluger 2011). People with ID, especially those with severe or profound ID can present with mobility and gross motor movement limitations. People can have difficulty conforming to positioning due to perhaps contractures and many people with ID can consequently be excluded from particular assessments based on their physical unsuitability (Kirschner *et al.,* 2007, Wilkinson *et al.,* 2007). Further exclusion can occur as a result of people's difficulties in compliance with the instruction for these assessments, culminating in these issues restricting the ability of people with ID using many diagnostic tools, for example the dual energy x-ray absorptiometry (DXA). Other barriers reported when accessing health services include inaccessible examination tables, waiting rooms or treatment clinics being too small for wheelchair access, inadequate parking or inaccessible ramps (Iezzoni *et al.,* 2000, Iezzoni *et al.,* 2010). The built environment still lacks functionality and accessibility despite many regulations the focus seems often to be on a stereotypical physical disability and other functional and cognitive impairment are disregarded.

3.3.4 Impairing attitudes

Regularly people with ID are dependent on healthcare by proxy. Very often people with ID may not be aware of the medical implications of symptoms they are experiencing, therefore their support person is a vital component for the smooth access and interpretation of their health needs. However, frequently family members or support carer's are not listened to (Turner and Robinson 2011) leading to discontinuity. Key issues to be overcome are identified by Krahn *et al.,* (2006) and include healthcare professional attitudes. If practitioners assume that because of their disability people with ID are less able, a situation can occur whereby the person with ID will be subject to lack of opportunistic screening. Instead of the process adapting to the person the person is viewed

as having to adapt to the process. Clinicians ought to ensure all citizens are offered the same standard of care, therefore, if the person meets the criteria for screening, screening should be adjusted and offered. Further, insufficient time is often assigned to an appointment resulting in a rushed experience. As a result health needs can go unmet (Krahn *et al.,* 2006). The poor experience at this one health visit can negatively impact on further and subsequent visits, culminating in reluctance by the person with ID to attend and engage in vital healthcare in the future. Conversely, the support person may lack the information or knowledge of the person with ID and this lack of information can impact on the care and support people with ID ought to receive when attending general healthcare setting appointments (Hatton *et al.,* 2011, Martínez-Leal *et al.,* 2011, Cooper *et al.,* 2004). Perry and colleagues note, in their review of health checks for people with ID, that some GP practices were reluctant to engage in the health checks of people with ID however, with the introduction of support and education, health check provision was improved (Perry *et al.,* 2008).

Very often there is a lack of medical or family history even if the person is living within a supported community house or residential setting. Martin (2003) attributes this gap in knowledge to frequent change in carers. This is of great concern in the current health climate in Ireland. The Irish healthcare profession is in flux with many senior staff retiring and many more emigrating (Burke 2015 , Cullen 2015) along with the fact that the ID services are experiencing great change (Health Service Executive 2011). People living within residential settings are now, due to needed reform and policy change, moving to more integrated settings in the community, accessing the community services and community general healthcare facilities (HSE 2011). A new model of health care delivery is envisaged for those moving into the community setting. People with ID will no longer exclusively access their health care through the service provider, but will engage with the local primary care team, community and public health services (HSE 2011). This vision is how it most definitely ought to be, however, public services require much work to advance equity of access for all citizens. Designing services with reasonable adjustment to cater for people with ID and promoting education for healthcare professionals contribute to advancing equal service provision. Improving understanding of the health needs of people with ID by including objective health assessments in research is one such contribution that can advance a goal of understanding health needs better, as well as providing empirical evidence of disease patterns. The opportunity to identify and measure undiagnosed disease and provide evidence to underpin policy and health promotion practice is imperative.

It can be seen from this brief overview of the literature people with ID present with increased risk of ill health which in turn can contribute to overall poor bone health. Preventing and delaying the consequences of poor bone health is possible. Early detection of osteoporosis is recommended utilising clinical risk factor findings, clinical history and x-ray evidence of osteopenia and osteoporosis (Health Service Executive (HSE) 2008). Brenner (2008) describes osteoporosis as the myocardial infarction of the orthopaedic world. Osteoporosis and subsequent fractures from falls are unnecessary. By implementing detection and assessment measures poor bone health can be addressed and reduced. However, people with ID have unique contributors that impact on their bones. Therefore, specific tools to assist with the detection of bone compromise among people with ID are required. Such findings support the prudence of investigating the area of bone health for this specific group. Building empirical evidence on the bone health of adults with ID is imperative. The inclusion of bone health investigations in the IDS-TILDA study offers a unique opportunity to pursue such research, and to contribute to the development of a body of knowledge that will support intervention strategies.

3.4 Conclusion

This chapter presented an overview of the risk factors and challenges associated with the development of poor bone health among people with ID. Examining the literature for the known risk factors with a specific focus on ID the researcher identified the specific conditions and factors that directly contribute to possibly an unrecognised high level of osteoporosis among this population. Although some risk factors are inevitable and unalterable those factors that are amenable require intervention to prevent a costly impact on the lives of people with ID. Barriers that increase the person's exclusion, impact on quality of life and contribute toward stigmatisation and disenfranchisement of people with ID were discussed. Inaccessible facilities, equipment and communication techniques continue to compromising the health care experience of many people with ID and leads to a paucity of understanding of their health needs. Improving and designing services with reasonable adjustment to cater for people with ID and promoting education for healthcare professionals will contribute to advancing equal service provision. To that end, the researcher has investigated the bone health among people with ID who are participants in a uniquely designed longitudinal study examining the lives of older adults with ID in Ireland namely, *The Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing*

(IDS-TILDA). The next chapter shall present an overview of the design, sampling, data collection and methods employed by the IDS-TILDA study.

CHAPTER 4

THE INTELLECTUAL DISABILITY SUPPLEMENT TO THE IRISH LONGITUDINAL STUDY ON AGEING (IDS-TILDA)

"Most of the important things in the world have been accomplished by people who had kept on trying when there seemed to be no hope at all"

- Dale Carnegie

Chapter summary

This chapter introduces the reader to the *Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing* (IDS-TILDA). This is the first time people with ID are represented in a nationally longitudinal study on ageing that is comparable to a national general longitudinal ageing study (TILDA). IDS-TILDA is designed to capture the unique and different health, social, living and community participation issues, and needs among adults over the age of 40 years in Ireland. It commenced in 2008 with a pilot study and now has two main study waves of data collection complete. Ethical approval for the study was granted by the University of Dublin, Trinity College Faculty of Health Sciences ethics committee at each wave, and by all 138 service providers involved in the IDS-TILDA study. The second wave included objective measures of health which also gained ethical approval at Wave 2 [2014]. IDS-TILDA has two robust frameworks underpinning the study. Firstly, the conceptual frame that encompasses all domains of health and living and an outline of this framework is presented in this chapter. Secondly, a values frame that guides the study, promotes the inclusion of the participants at all stages from inception to dissemination and guides the interviewers, the development committees and study researchers in their approach to researching with people with ID. Being conscious of ethical responsibility the ethical guiding principles of the IDS-TILDA study are presented. As this researcher had sole responsibility for the *Health Fair* data collection and data the researcher elucidates the application of the ethical principles in this chapter.

4.0 Introduction

The IDS-TILDA study is a national longitudinal study on ageing and ID. The study commenced in 2008 following in the footsteps of the general population study '*The Irish Longitudinal Study on Ageing*' (TILDA) (Kenny *et al.*, 2010). The TILDA study commenced in 2006, at that time there was no national longitudinal study on ageing in Ireland to provide accurate health and socio-economic data on growing older in Ireland, essential information to inform policy and strategic forward planning for a growing ageing population. A nationally representative sample of adults over the age of 50 years was selected from the GeoDirectory (Barrett *et al.*, 2011). Unintentionally the sampling frame and participant recruitment process chosen by TILDA excluded many people with ID as their criteria required all participants to self-consent and only those living independently at home were recruited. Also in their first wave of data collection, TILDA did not recruit from any residential or nursing home setting (Kenny *et al.*, 2010). The opportunity was therefore present to develop an ID specific study harmonised with TILDA, from this IDS-TILDA was established.

The uniqueness of the IDS-TILDA study is that it is the first of its kind that is directly comparable to a longitudinal general population ageing study.

4.1 IDS-TILDA design and recruitment

Ireland was in the unique position for such a study as it has a national intellectual disability database (NIDD) of people with ID who are in receipt of ID services. From this database random selection of participants stratified by age, gender, level of ID and living circumstances was possible. The IDS-TILDA study commenced in 2008 and a pilot feasibility study was conducted (McCarron *et al.*, 2009).

The first wave of the main study commenced in 2009. The inclusion criteria for the study stated that, people had to be registered on the NIDD of Ireland and be aged 40 years or over. Participants were randomly selected from the NIDD. To maintain anonymity of potential participants during the selection process a gate keeping system, in the form of the database administrators of each region in the country was used. The NIDD selected 1800 PINs, 1600 of which were released to the regional database co-ordinators in incremental stages on instruction of the IDS-TILDA team. This was to facilitate the steady and ongoing flow of data collection. Each database co-ordinator

attended an educational seminar regards their role and responsibility. The IDS-TILDA team prepared invitation packs (see appendix 4) which were sent to the database coordinator. The database coordinators then addressed and forwarded the invitation packs to the individuals. Full details of the sampling, recruitment and participation process of Wave's 1 & 2 can be seen in *figure 4.0.*

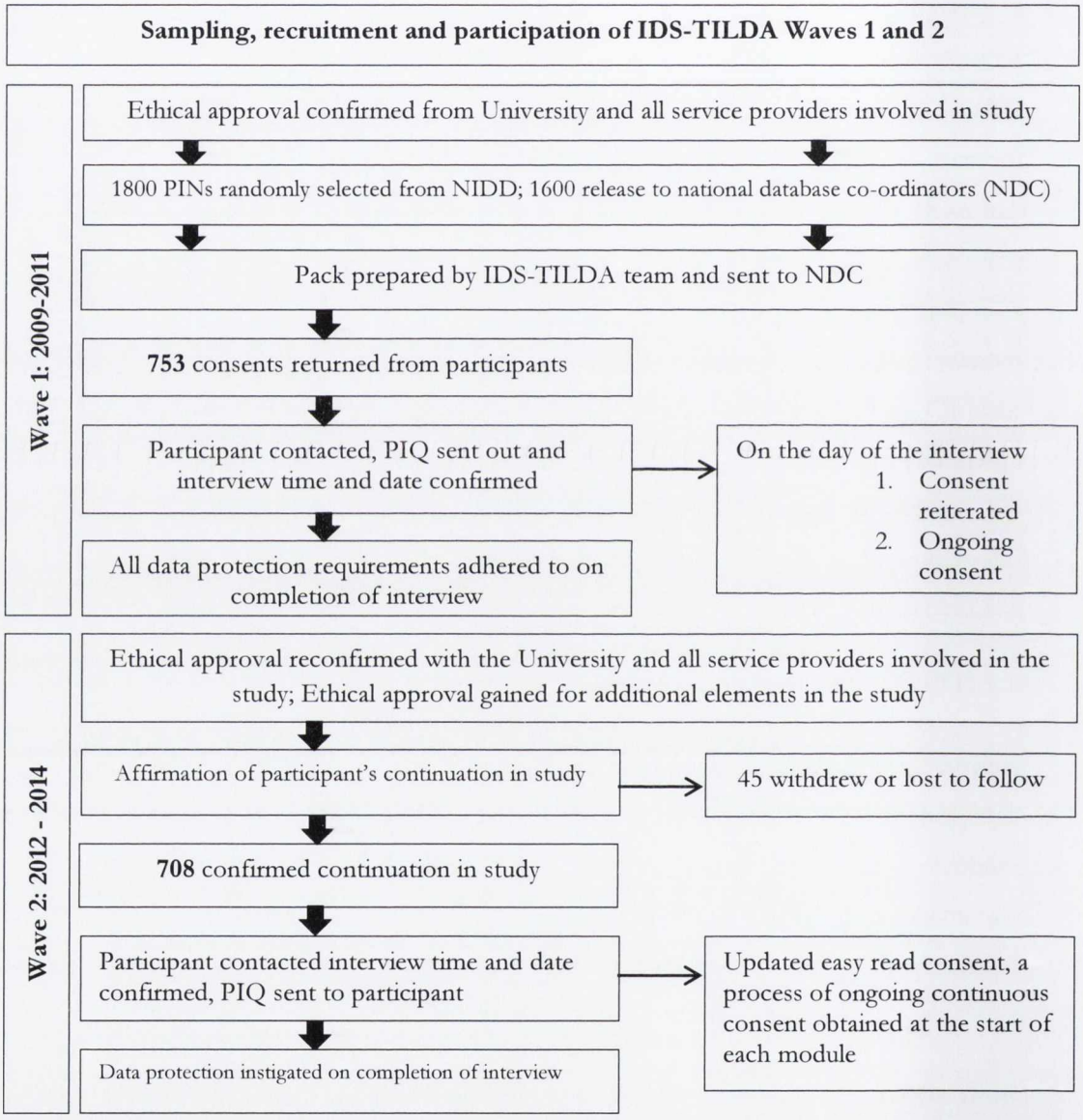


Figure 4.0 Sampling, recruitment and interview process for IDS-TILDA Wave's 1 and 2

To fully promote the inclusion of people with all levels of ID, all material sent was in an "easy-to-read" format emphasising plain language and pictorial representation (compiled and reviewed in collaboration with a number of advocacy groups throughout Ireland). The invitation pack consisted of an easy-to-read cover letter and information booklet on the study, an easy-to-read consent form, stamped addressed envelope and a family information pack. The purpose of the family information pack was to inform the participant's families of the study and provide them with the opportunity to support their family member making the decision to take part or not. To indicate their wish to participate, each selected person was invited to return their easy-to-read written consent. On receipt of this consent the IDS-TILDA team became aware of the person who wished to take part in the study and the participant was allocated to a trained field researcher. The returned consents were assigned an anonymous IDS-TILDA study PIN. All data protection procedures were instigated to maintain the participant's privacy and to that end all identifiers are kept in a locked cabinet within a locked facility separate from any information identifiable by PIN.

Crucial to the design of the study was the Scientific Advisory Steering Committee and the Advocacy Groups (People with ID who acted as independent reviewers and advisors). These groups collaborated closely with the IDS-TILDA team through protocol, process and supporting material development, contributing to and influencing the underpinning conceptual framework (see *figure 4.1*) and the values framework (see *figure 4.2*) which benchmarks and guides the study.

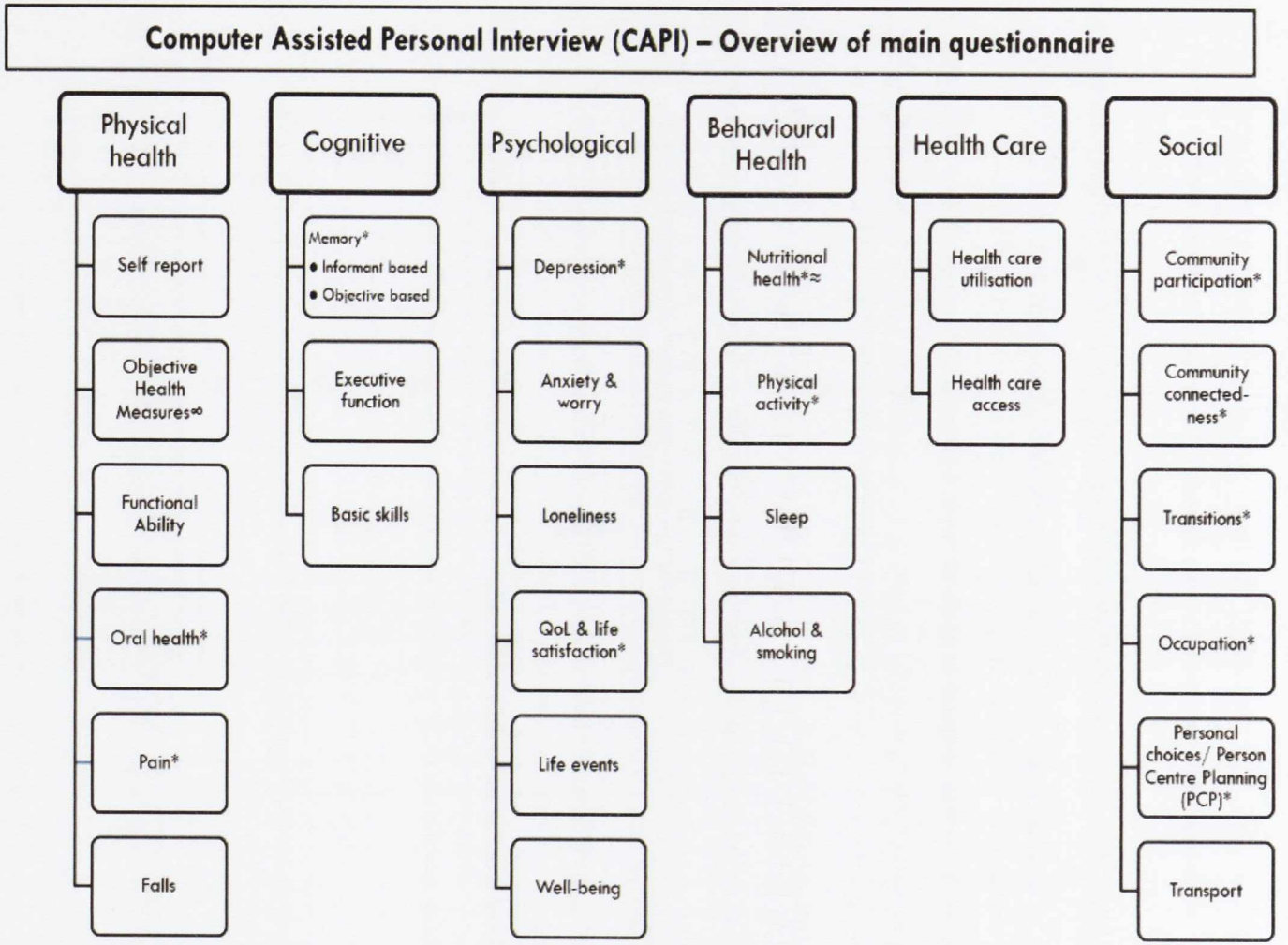


Figure 4.1 Conceptual frame of the Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing

Source: *Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing*

The values frame (*figure 4.2*) underpins the IDS-TILDA study and has guided the development and review of the questionnaire, training of the field researchers, interviewing technique and dissemination strategy of the study. The values frame is integral to the conduct of the study and represents the principles the study values.

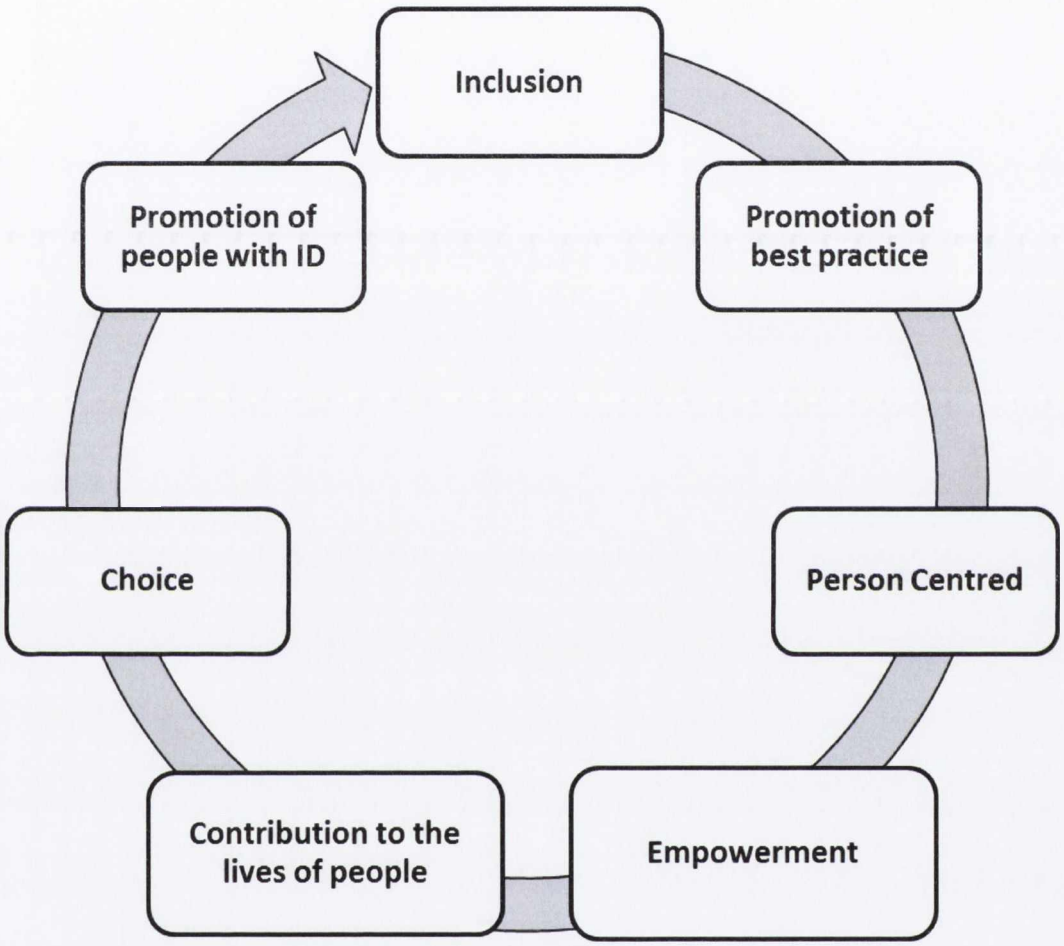


Figure 4.2 Values frame of the Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing

Source: *Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing*

The IDS-TILDA team have a robust '*keeping in touch strategy*' (see appendix 5) to maintain contact with participants, keep them up to date on the study and to maintain accurate database information on participant location. Through this system participants are encouraged to relay any changes in their living circumstances therefore making contact for future study waves more convenient. On commencement of Wave 2 [2014] no sample refreshment was pursued and all consented participants from Wave 1 [2011] were contacted and invited to continue in the study. Each participant received an updated information pack and consent was reaffirmed at the time of the face-to-face interview (see appendix 6 for Wave 2 consent).

4.2 IDS-TILDA data collection Wave 1 and Wave 2

Each field researcher underwent robust interview and protocol training prior to entering the field which included ethical, IT, communication and safety procedure instruction.

Each field researcher received a structured comprehensive training program on the administration of the overall protocol and the field researcher 'shadowed' an experienced interviewer to enhance the standardisation of the face-to-face interview. The assigned field researcher telephoned the potential participant to inform them of the study process and arrange the face-to-face interview. A pre-interview questionnaire (PIQ; *see figure 4.3*), a self-report document, was sent to the participant a minimum of one week in advance of the face-to-face interview. This was to facilitate the participant obtaining assistance to complete the questionnaire and time to gather the information about the questions which comprised mainly of self-reported doctor's diagnosis of health conditions, medications and health care utilisation. On the day of the face-to-face interview a computer assisted personal interviewing (CAPI) system was used (IBM 2011a) and the interview took approximately 90 minutes to complete.

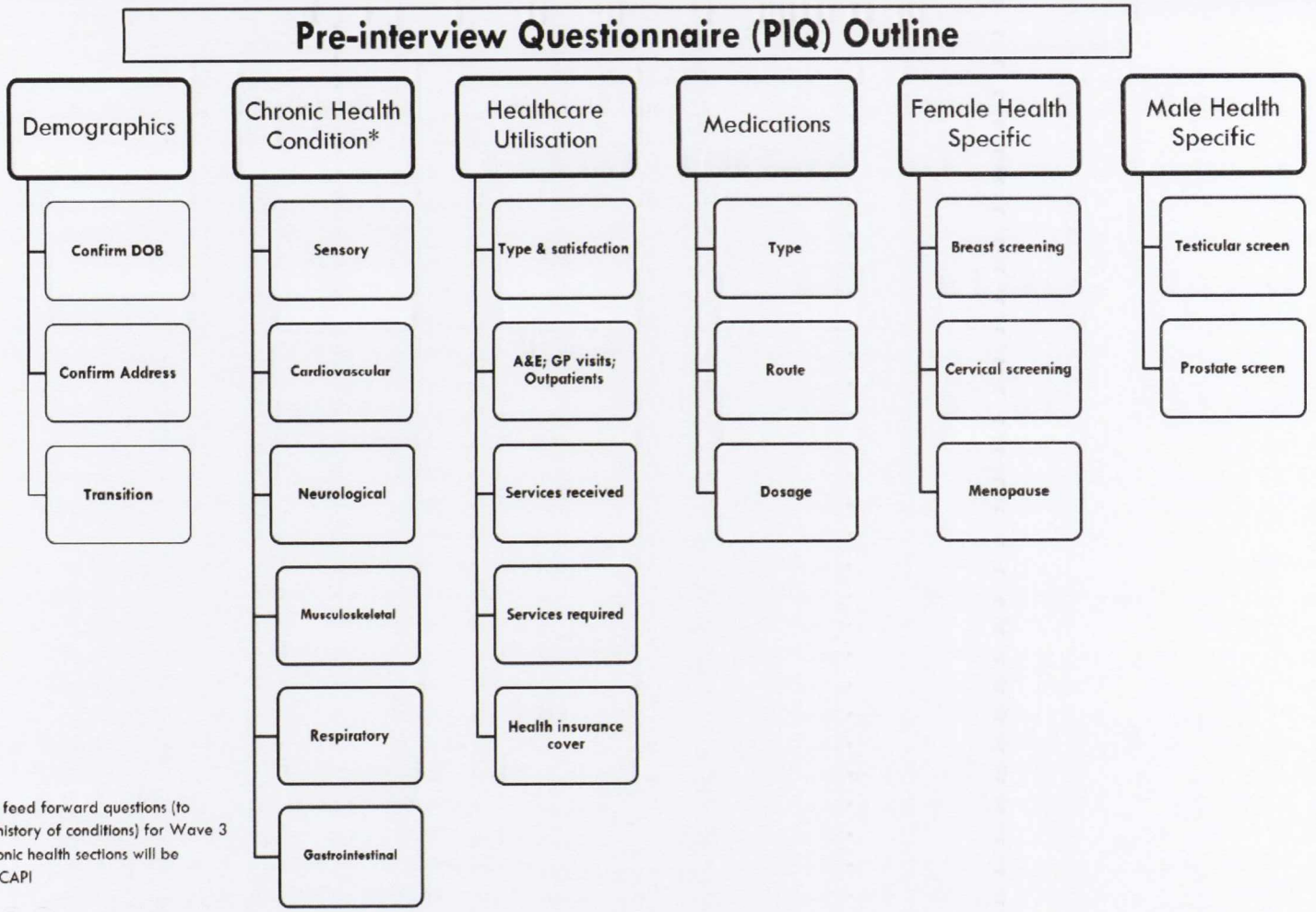


Figure 4.3: Pre-interview questionnaire modules of the Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing

Source: *The Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing*

4.2.1 Interview styles, review and the inclusion of objective measurement

There were three styles of interview offered and this was determined by the participant's level of communication and level of ID.

1. A self-report only interview: The participant answered all their interview questions independently.
2. An interview supported by a proxy: The proxy (carer or keyworker) supported the participant with answering questions; this could be in the form of interpreting for the interviewer or explaining to the participant, in language familiar to them, the content of the question.
3. Proxy only interview: The interview was totally completed by a proxy on behalf of the person with ID however for this style of interview some modules were omitted as they were self-report only for example ageing perceptions, some of the mental health questions and those questions that referred to how the participant felt.

The proxy was a person who knew the participant for a minimum of 6 months. The questions for the CAPI interview are structured through a number of modules (*see figure 4.1*) with each addressing demographic characteristics; cognitive health maintenance; happiness; physical health; activities of daily living and instrumental activities of daily living and related support needed; mental health; occupation; lifelong learning; day services; behavioural health; social participation; social connectedness; life satisfaction; ageing perceptions; personal choices; and sources of income.

Between Wave's 1 and 2 the overall protocol was reviewed and modified. Some items were rested for Wave 2, some items were removed and additional items were included based on participant feedback and emerging results. For further details on questions removed, rested and included see Carroll *et al.,* (2014). On consideration of the ethical, financial and participant burden and in collaboration with the Scientific Advisory Steering Committee, unlike the general population study TILDA, IDS-TILDA decided not to include objective measures in the first Wave of the study. Following conclusion of Wave 1 and driven by its success, the willingness of the participants and the increased focus on successful active ageing, objective health measurements (henceforth referred to as the *Health Fair*) were confirmed and included as part of Wave 2. The *Health Fair* was conducted separately to the main IDS-TILDA Wave 2 study and ran approximately three months behind the main study. The field researchers of the main study introduced the *Health Fair* to participants informing them that it was coming and distributing the

Information Poster (see appendix 7). The Health Fair consisted of a suite of 8 non-invasive measures (*see figure 4.4*) chosen and designed to specifically promote comparison with TILDA. These included height and weight, waist to hip ratio, blood pressure (both sitting and standing), Timed Up and Go, grip strength and bone density measurement performed by calcaneus quantitative ultrasound (QUS). The use of the MUST tool to provide alternate surrogate measures, ulna length for height and mid upper arm circumference for estimation of body mass index (BMI), were included for people who would be unable to engage in height and weight assessment (see *Health Fair Protocol* appendix 8). Following consultation with the advocacy groups and the Scientific Advisory Committee the *Health Fair* protocol was reviewed and defined. The roll out of the *Health Fair* was conducted through a three phase inclusive approach in conjunction with the participants, their family members or carer's and the service providers where they attended for services (see appendix 9) for detail of the process for setting up, contacting and completing the *Health Fair*). More detailed information on the *Health Fair* are presented in Chapter 7.

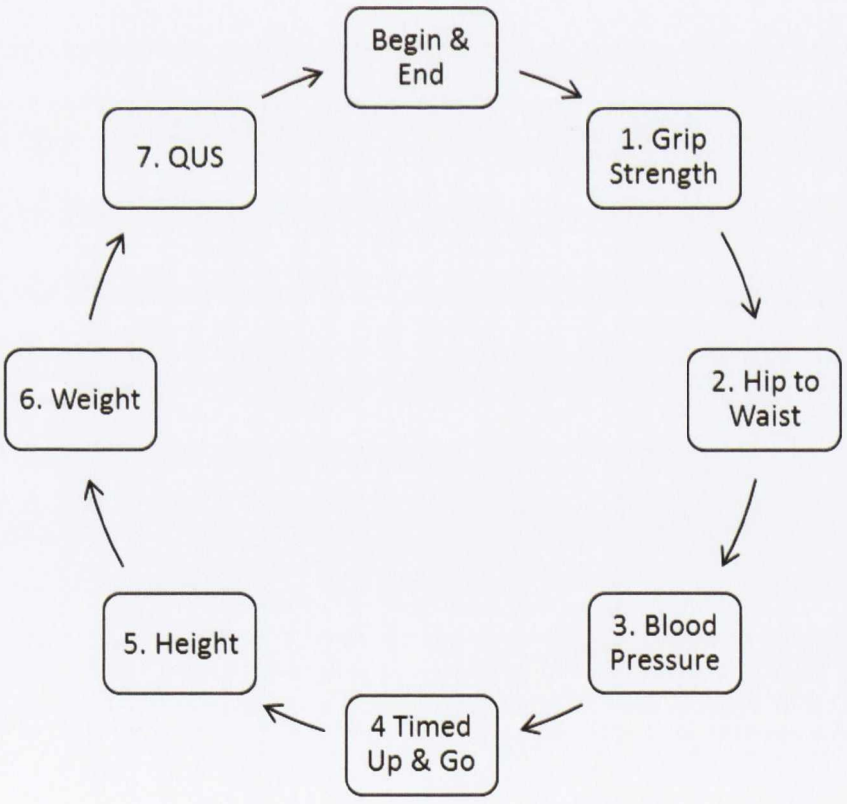


Figure 4.4 Health Assessment: A suite of 8 health assessments each purposely positioned to lessen the degree of inconvenience for the participant.

4.3 Examining medication data

In both Wave 1 and Wave 2 of the IDS-TILDA study participants/proxy were asked to record in the pre-interview questionnaire their current medications and supplements (including prescribed or over the counter medicines) Medicines were recorded by brand/generic name, including prescription and non-prescription, over the counter, herbal and alternative medicines and any food supplements taken, and where available the dose and frequency. Medication use was defined as regular use (every day or every week), this included oral, parenteral, topical, and ophthalmological and inhaled medicinal products. The accuracy of this information was further improved by cross checking the medication information in the face to face interview (O'Dwyer 2015).

4.3.1 Classification of medicines

For the purposes of this thesis, medications and supplements were recorded using the World Health Organisation Anatomical Therapeutic Chemical Classification (ATC) classification code, International Non-Proprietary Name (INN), and brand name where available (WHO 2011a). See **table 4.1** for the ATC codes used in this thesis. The ATC system is an internationally used system to code medications, controlled by the World Health Organisation Collaborating Centre for Drug Statistics Methodology. The ATC system classifies drugs into different groups according to the system or organ on which they act and/or their chemical or therapeutic characteristics. Drugs are classified in groups at five levels. Drugs are classified into 14 main first level groups, with one therapeutic or pharmacological second level subgroup. The third and fourth levels then consist of the chemical/pharmacological or therapeutic subgroups, and the fifth level consists of the chemical substance.

Table 4.1 ATC Codes used in thesis

Medication	ATC Code
Antipsychotic agents	N05A
Antidepressants	NO6A
Anxiolytics	N05B
Sedative/hypnotics	N05C
Anti-epileptics	N03A

4.4 IDS-TILDA distribution and participation

In total, 753 participants from 138 service providers throughout the country took part in Wave 1, representing 8.9% of the total registered population aged over 40 years. Overall, 55% (n=415) were female and 45%, (n=338) were male. All levels of ID were represented with the highest percentage (44%, n=346) within the moderate ID range (McCarron *et al.*, 2011).The same participants were invited to participate in Wave 2 and a total of 708 reaffirmed and took part, see **table 4.2** for participant demographics of Wave 1 and Wave 2.

Table 4.2: Demographics of participants Wave 1 and Wave 2

Data Collection Waves*	Demographic	*n*	%	Totals
Wave 1				**753**
Gender	Male	336	44.8	
	Female	417	55.2	
Age	40 – 49	274	36.4	
	50 – 64	344	45.7	
	65	134	17.8	
Level of Intellectual Disability	Mild	166	23.9	
	Moderate	323	46.5	
	Severe & profound	206	29.6	
Living Circumstance	Independent/Family	129	17.1	
	Community group home	268	35.6	
	Residential setting	356	47.3	
Wave 2				**708**
Gender	Male	312	44.1	
	Female	396	55.9	
Age	43 – 49	198	28.2	
	50 – 64	355	50.6	
	65	148	21.1	
Level of Intellectual Disability	Mild	154	23.8	
	Moderate	301	46.5	
	Severe & profound	192	29.7	
Living Circumstance	Independent/Family	113	16.2	
	Community group home	303	43.3	
	Residential setting	283	40.5	

**Missing obs: Not all participants provided data for all questions asked.*

4.5 Ethical Considerations

> *Research with vulnerable populations challenges us to consider once again ethical principles basic to research. Issues of providing informed consent, maintaining confidentiality and privacy, weighing the risks and benefits of a study and paying attention to issues of fairness are all especially important with working with groups who are vulnerable.*
>
> (Flaskerud and Winslow 1998: 69)

Fundamental to all research studies is the consideration of ethical principles. Ethical consideration does not stop with the granting of ethical approval the researcher must be continuously conscious of ethical principles which will provide guidance in the ethical dilemmas researchers face. These principles are the guiding principles that underpin the IDS-TILDA study. This researcher acknowledged and considered these principles carefully ensuring to apply additional dimensions where necessary for example, informed consent. The researcher observed at all times and in accordance with An Bord Altrainais (ABA) ethical research guidelines, to the *Code of Professional Conduct and Ethics of each Nurse and Midwife* (An Bord Altranais 2014). The primary data collected by the researcher was pertaining to health assessments. All ethical principles apply to the collection of the Wave 1 & 2 IDS-TILDA data and in Wave 2 the sole responsibility for the health assessments (objective measures) data collection was held by this researcher. The following are the guiding ethical principles applied throughout her study.

Guiding ethical principles

- Attaining ethical approval
- Respect for persons/autonomy
- Informed Consent
- Anonymity and confidentiality
- Beneficence
- Non-maleficence
- Justice/fairness and veracity

(*Beauchamp and Childress 2001*)

4.5.1 Ethical Application and Attaining Approval

Ethical application to undertake the objective measures was included in the Wave 2 IDS-TILDA study submitted January 2013. All service providers were furnished with the renewed ethics application highlighting the additional items, the items removed and the rationale for each amendment. The inclusion of the objective measures was a new additional item and the rationale and any ethical implications were highlighted and explained. Ethical approval was granted by the University Faculty of Health Sciences for Wave 2 of the IDS-TILDA study which included the objective measures section (see appendix 10). Each service provider then reaffirmed ethics following this review and approval. Ethical approval from each of the services is not included in the appendices of this report firstly as there are a number of documents which would prove too cumbersome and secondly it would serve to identify individual service providers and in turn the potential of identifying individual participants which would be in breach of data protection protocols and procedures held by the IDS-TILDA study.

4.5.2 Respect for Persons

The capacity for the participant to act without direction and to decide without interference promotes respect for persons. Whilst people with ID are considered to be a vulnerable population the researcher was conscious not to impose vulnerability. Co-production, the researcher exacerbating the vulnerability of the individual, poses to disempower therefore through the adoption of an emancipatory approach, the objective measures procedures design that reinforced the person's achievements and the promotion of positive understanding of all activities contributed to the promotion of respect for persons. At all stages of the *Health Fair*, from initial phone contact to the day of the *Health Fair*, the rights of the person were considered the highest priority. To that end, it was imperative that all participants were offered the choice to participate or not. Additionally, it was made explicit to each participants that the results sheet the researcher retained did not have their name on it, at no time would the name and assigned PIN be available to any parties save the IDS-TILDA research team, all soft and hard copies were stored in secure encrypted locations in accordance with the data protection procedures of the study and the Data Protection Act (Government of Ireland 2003), therefore maintaining the person's anonymity and confidentiality.

4.5.3 Informed Consent

Informed consent is a process that ensures all participants understand the potential risks and benefits of taking part in the study. The four elements of informed consent according to Burns and Grove (2010) include disclosure of all crucial information relating to the study, presenting the information in such a way that the participant will gain full understanding, the capability of the participant to give consent to take part and the consent is given without coercion or pressure to do so and is fully voluntary. In addressing these issues the researcher has at every stage of data collection provided easy-read information in a timely fashion so as to enable the participant seek support where required, has reinforced the voluntary nature of the participants inclusion, has obtained consent from the participant before doing any interview or assessment ensuring the participants rights are elucidated, has sought support from familiar carers where communications difficulties have arisen and has reiterated consent in an ongoing fashion before every assessment was carried out. The implementation of an emancipatory process also lends itself to bringing informed consent to the fore as the researcher is guided by the participant as to how much they want to be involved (see appendix 11 for consent form utilised in the *Health Fair*).

4.5.4 Anonymity and Confidentiality

The researcher assured all participants of the PIN system the IDS-TILDA study has in place. All data entered for analysis is anonymised using this PIN system. Participants are reassured that at no time do individual names or service provider names appear or be identified in any report or publication written or published from the research. All computers used for data entry are double encrypted and all data stored is done so on a secure drive hosted by the university – all data entered by the researcher was transferred to this secure server as soon as was possible after entry. At no time was the data available to any person with the exception of the IDS-TILDA research team. Assurance of this was advised to each participant. Being cognisant of the Data Protection Act (Government of Ireland 2003) all hard copies of data collected are stored in locked cabinets in a locked facility and shall be kept for a stipulated number of 5 years post the completion of the main parent study (IDS-TILDA) after which time they will be destroyed.

4.5.5 Beneficence and non-maleficence

These principles impose a duty to maximise benefit and minimise harm for all participant in a research study (Polit and Beck 2008). In accordance with ethical approval all participants received their results of the assessments on the day of completing the *Health Fair* (see appendix 12). The results were explained and the participants were encouraged to talk with their carer or support person. Any abnormal findings were highlighted and the participant was advised to seek further assistance in the form of their general practitioner (GP) where necessary. This was marked clearly on the results card the participant took home. The researcher's contact details were also available on the card should they need to make further queries. It was also stressed that this research is part of the larger parent study, IDS-TILDA, and as such will contribute to the development of the overall picture of how people with ID grow older in Ireland. Furthermore, it is hoped that the findings may benefit and influence changes in clinical practice, at academic level and at policy level, in the future.

4.6 Conclusion

This chapter has introduced the reader to the IDS-TILDA study, its design, sampling and recruitment. The underpinning conceptual frame along with the values frame that guides the IDS-TILDA study has been presented. The questionnaire modules included in both Wave's 1 & 2 are highlighted and the process of data collection of the main IDS-TILDA study has been presented. The researcher has provided a brief overview of the interview styles and approach of the IDS-TILDA study, identified the drivers and influences of including objective measures in the second Wave [2014]. To that end, the researcher has given a brief introduction of the health assessments in Wave 2 however this will be presented in more detail in Chapter 7. An overview of the participation and demographics of the IDS-TILDA study is also presented. And in conclusion, focusing on the *Health Fair* the researcher focuses and elucidates the ethical principles applied to that data collection process.

The particular area of interest to this thesis is osteoporosis. Building a comprehensive picture of the bone status of adult with ID would provide a rich source of data to help understand the complex relationship between physical and environmental factors and pre-existing impairment impacting on the ageing of people with ID. Objective health assessments in research help advance this goal by providing empirical evidence of

disease patterns, identifying undiagnosed disease and developing an evidence-based approach to disease management and health promotion practice. The opportunity to identify prevalence of osteoporosis and rates of risk factors among people with ID in Ireland through the IDS-TILDA study affords an opportunity to contribute to the amelioration of this debilitating silent disease. To that end the researcher firstly establishes the extent of osteoporosis among the IDS-TILDA cohort through the exploration of the first wave [Wave 1, 2011] of data collection. This will provide information on the level of diagnosis that already exists among people with ID in Ireland and elucidate a rationale for the inclusion of objective measurement of bone health in the second wave of the overall IDS-TILDA study. The baseline picture of the bone health of older adults with ID in Ireland is presented in the next chapter.

CHAPTER 5

AN EXPLORATION OF THE BONE HEALTH OF OLDER ADULTS WITH AN INTELLECTUAL DISABILITY IN IRELAND - RESULTS FROM THE FIRST WAVE OF IDS-TILDA

"Small opportunities are often the beginning of great enterprises."

Demosthenes: Ancient Greek statesman

Chapter summary

This paper presents the findings on the bone health of older adults participating in the first wave of the *Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing* (IDS-TILDA). Following an examination of the literature a risk factor framework was developed and mapped to the data from the IDS-TILDA study through which the investigation of the bone health of the participants was guided and explored. Overall 8.1% (n=81/753) self-reported a doctor's diagnosis of osteoporosis. Of those who replied to the screening question, 11.1% (n=43/389) had DXA (Dual energy X-ray Absorptiometry) within the previous 2 years and overall 17.8% (n=134/753) were taking calcium and vitamin D supplementation. However 38.2% (n=287/753) were taking antiepileptic medication, over 57% (n=238/412) of the females had experienced the menopause, physical inactivity was high (70.3%, n=528/753) and over 60% (n=174/584) were overweight or obese. There was a moderately high level of reported history of fractures 20.6% (n=152/738) and of those who fractured there was very little difference between genders with males experiencing slightly more fractures than females, 20.7% versus 20.5% respectively. As would be expected fracture was strongly associated with a history of falls ($p<0.0001$), epilepsy ($p<0.0001$), having difficulty walking ($p<0.0001$) and having a self-reported doctor's diagnosis of osteoporosis ($p<0.008$). The strongest predictors identified for a diagnosis of osteoporosis were, being over the age of 65 years (OR 6.531, CI02.823-15.110, $p<0.0001$), being female (OR 4.582, CI2.289-9.172, $p<0.0001$) or have a self-reported doctor's diagnosis of arthritis (OR4.073, CI2.217-7.483, $p<0.0001$). Considering the literature on Down syndrome (DS) it is interesting that DS did not emerge as a risk factor. This could possibly be due to the fact that the numbers within the sample were small (n=147) and people with DS may require separate investigation. Nevertheless, the level of risks for poor bone integrity were high and the rates of screening were low. Bearing in mind the insidious nature of osteoporosis, it is safe to assume that the self-reported level of doctor's diagnosis may not capture the possible levels of osteoporosis that exist within this cohort. This paper is based on doctors diagnosis and further objective measurement investigation is warranted.

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5.0 Introduction

Osteoporosis is described as a multifactorial disorder characterised by low bone mass and micro-architectural deterioration of bone tissue leading to enhanced bone fragility and a consequent increase in fracture risk (WHO 1994; Kanis *et al.,* 2013; Cooper 2014). The insidious nature of the condition leads to under diagnosis, often only detected post clinical fracture (Kanis *et al.,* 2013; Kanis 2002). Internationally the World Health Organisation's (WHO) operational definition of osteoporosis is well established and utilised for over 20 years (WHO 1994). The presence of osteoporosis is defined as having a bone mineral density (BMD) value 2.5 standard deviation (SD) or more below the young female adult mean, with severe osteoporosis described as above in the presence of 1 or more fragility fractures. BMD is measured by dual energy x-ray absorptiometry (DXA) which is the 'gold standard' for diagnosis of osteoporosis, evaluation of fracture risk and monitoring skeletal changes (Kanis *et al.,* 2008b, Kanis *et al.,* 2009a). More recently alternate BMD measurement devices have been introduced and the International Society for Clinical Densitometry has confirmed that these peripheral bone density measurement devices have value for assessing fracture risk and identifying people who may require further diagnostics with DXA. One such technique is the quantitative ultrasound (QUS) which is a quick, non-invasive method of scanning the os calcis (heel), a highly trabecular and weight bearing bone similar to the hip and spine, the sites commonly utilised for DXA.

There have been a plethora of studies among the general population describing the pathogenesis and impact of osteoporosis however there are limited investigations describing prevalence among adults with intellectual disabilities (ID). It is known that people with ID experience higher levels of adverse health conditions than people without ID (vanSchrojenstein Lantaman-De Valk *et al.,* 2000). Indeed McCarron *et al.,* (2013a) identified higher prevalence of multimorbidity and complex conditions in comparison to the general population. Very often health needs go unmet and unrecognised (Lennox and Kerr 1997, Beange and Durvasula 2001; Cooper *et al.,* 2004; Kerr 2004; Emerson and Hatton 2013). One such area quite often neglected or omitted on physical health checks is bone health with preventative services also underutilised (Lewis *et al.,* 2002, Michael 2008, Srikanth *et al.,* 2011).

5.1 Exploring risk factors

Ageing is a well-documented contributor to bone loss (Datta *et al.*, 2008) and until recently longevity in persons with ID was lower than the general population. These trends are changing; Kelly and O'Donohoe (2014) describe an increase from 29% in 1974 to over 48% in 2013 among people with ID over the age of 35 years. However it is also important to note that osteoporosis is not always the result of accelerated bone loss due to ageing. Peak bone mass is attained in early adulthood and remains constant into the mid to late thirties. Nonetheless if an individual does not reach optimal bone mass, as is the case for people with Down syndrome (DS), there is an increased risk of developing osteoporosis (Hayes and Batshaw 1993).

Osteoporosis occurs in both genders however women are at higher risk of osteoporosis and related fractures due to such factors as lower muscle mass, fluctuation of oestrogen levels and menopause (Melton 2003). Some females with ID and especially those with DS have added risk loading due to late menarche and early menopause (Knickmeyer *et al.*, 2006, van Schrojenstein Lantman-deValk *et al.*, 2002, Carr and Hollins 1995). It is also recognised that hypogonadism associated with reduction in pubertal growth spurt thus low oestrogen and testosterone levels is prevalent among males with DS (Seeman *et al.*, 1983, Hsiang *et al.*, 1987, Schuf *et al.*, 1997).

Level of ID can have adverse effect on bone health as those with more severe to profound ID are more likely to present with increased co-morbidities (McCarron *et al.*, 2013). Non-ambulatory status has been correlated to higher rates of osteoporosis and fracture (Tyler *et al.*, 2000, Baptista *et al.*, 2005). Also of note is that this cohort is more likely to experience fracture due to the higher incidence of epilepsy, subsequent antiepileptic drug use, and falls (Glick *et al.*, 2005). Communication challenges are also prevalent among adults with ID. These challenges make it difficult for people to express their own health needs, discomfort or pain which in turn can contribute to osteoporotic fracture going undiagnosed. Communication difficulties, especially with comprehension, also contribute to challenges for the person with ID understanding health screening procedures such as DXA. People with ID are very often unfamiliar with DXA and compliance with the specific instructions required for successful scanning do pose problems especially for those with severe or profound ID.

Syndrome specific risks of poor bone health have also been recognised (Angelopoulou *et al.,* 1999, Geijer *et al.,* 2014). People with DS are particularly at risk and indeed Angelopoulou *et al.,* (1999, 2000) has cited DS as an independent contributory risk factor. People with DS have higher prevalence of endocrine disorders such as hypothyroidism (Jaffe *et al.,* 2005). Karlsson *et al.,* (1998) in their longitudinal study identified a 33% development of hypothyroidism before the age of 25 years which in turn can contribute to poor bone health. McCarron *et al.,* (2014) reported that 28% of adults with DS over the age of 40 years had hypothyroidism. As people with DS age the prevalence of Alzheimer dementia increases rapidly (McCarron *et al.,* 2005), the associated epilepsy, increased risk to wander and fall, medications and increased difficulty swallowing combined with the already underlying risks predispose this cohort to further bone compromise.

In addition to complex health conditions lifestyle factors are also notable contributors to poor bone health. Bone requires stimulation to promote bone turnover, physical inactivity is notable among adults with ID and at a level that does not accrue health benefit (Robertson *et al.,* 2000, Beange *et al.,* 1999, McCarron *et al.,* 2011). Leslie *et al.,* (2009) confirm an association with immobility and osteoporosis in adults with ID which is further supported by Wagemans *et al.,* (1998). They note that as a consequence of poor mobility more than 75% of the cohort had poor bone density despite vitamin D supplementation as a preventative measure. Other contributing behavioural factors include poor dietary habits which contribute to an overall imbalanced diet and subsequently inadequate vitamin and mineral intake. Poor dietary habits can lead to gastrointestinal disturbance and coupled with low levels of exercise, lead to chronic constipation, this in turn effects bone health as chronic constipation can hinder the absorption of vitamins and minerals especially vitamin K in the large bowel, which is crucial in the synthesis of osteocalcin, an element essential in bone mineralisation and calcium ion homeostasis which is pro-osteoblastic in nature (Weber 2001, Pearson 2007). Further underweight or overweight status is also a reflection of imbalanced diet. Many of these states are prevalent among people with ID (Rimmer *et al.,* 2010, Morad *et al.,* 2007, Rimmer *et al.,* 2007, Yamaki, 2005, Emerson 2005).

5.2 Consequence of osteoporosis

Osteoporosis is often referred to as the silent thief of bone. The bone demineralisation weakens bone asymptomatically predisposing the bone to an increased risk of fracture which is the most common outcome of osteoporosis. The most frequently occurring associated fractures are proximal femur, distal radius or vertebral fracture, leading to pain, increased morbidity and loss of independence. Zimmerman *et al.,* (1998) reports of the high prevalence of hip fracture among residential settings for the elderly and how this impacts on the quality of life of those who fracture. In fact, Gold *et al.,* (1996) reports that 50% of those over 60 years who present with hip fracture will not regain full independence in their daily activities of living such as washing, dressing or walking unaided.

People with ID are at increased risk for poor bone health and although some risks are inevitable and unalterable, there are others which may be preventable. The aim of this paper is to identify the prevalence of and associated risk factors for osteoporosis among older adults with ID in Ireland. This opportunity is made possible through the use of the data collected by the Intellectual Disability Supplement to the Irish Longitudinal Study on Ageing (IDS-TILDA).

5.3 Methods

5.3.1 Study design and participants

Data was drawn from the first wave of the IDS-TILDA study. This study is a national longitudinal study on the health and well-being of adults with an ID as they age. In summary, 753 participants aged 40 years and over, with varying levels of ID, across a range of living circumstances were randomly selected from the National Intellectual Disability Database (Kelly *et al.,* 2009). IDS-TILDA is unique insofar as it is harmonised with the Irish Longitudinal Study on Ageing (TILDA) the larger study investigating ageing in the general Irish population (Kenny *et al.,* 2010). The protocol development was informed by a scientific committee and advocacy groups of people with ID, who reviewed and contributed to questions, amendments and, the development of easy-read material to assist in understanding the overall study, assist with obtaining consent and understanding the questions. For detailed study methodology, see McCarron *et al.,* (2011).

5.3.2 Ethical considerations

Ethical approval for the study was obtained from the Faculty of Health Sciences, Trinity College Dublin and from all the service providers involved in the study. The initial information pack sent to the participants included easy-read information, an easy-read consent and a family support package. A gatekeeper system was employed to preserve the participant's anonymity and only on return of the signed consent form did participant names become known to the study. All data collected is stored in adherence with data protection regulations (Government of Ireland 2003).

5.3.3 Data collection

Data was collected using a pre-interview questionnaire (PIQ) and a face-to-face interview. The PIQ was posted to the participants a week in advance of their face-to-face interview. This facilitated the participant having time to collect the required information and gain support for completion if required. The self-reported questions on doctor's diagnosis, medication use and healthcare utilisation were within the PIQ. The face-to-face interview utilised computer assisted interviewing on encrypted laptops. The data collected included a number of modules encompassing, dietary intake and frequency, activity levels along with social connections and community participation, activities of daily living, mental health and psychological well-being. The participant could complete the interview independently, be supported by a key worker or proxy or have the proxy complete the interview on their behalf. The proxy had to have known the participant for at least 6 months or more. In this study, 40.4% completed the face to face interview independently, 20.8% completed with support and 38.8% of the interviews were completed by proxy on behalf of the participants.

5.3.4 Statistical Analysis

For the purposes of this paper statistical analysis was conducted using the Statistical Package for Social Sciences (SPSS) version 22 (IBM 2011a). Preliminary analyses were initially carried out which included frequencies, distribution and identification of missing values. With consideration to missing data valid percentages only are presented throughout the paper. Descriptive statistics were used to analyse the demographic data and independent variables were explored stratified by gender, age, level of ID and living circumstance. In keeping with the theoretical frame presented below (*figure* 5.0) all variables were tested for significance however only significant values are presented. A *p*-value of

0.05 was considered statistically significant. Bivariate correlation for self-reported doctor's diagnosis of osteoporosis was calculated with Pearson's coefficient of correlation. Overall relationship between the dependent variable and the statistically significant risk factors and crude odds ratios are explored and identified using binary logistic regression analysis.

Identifying risk factors for poor bone health

Taking into consideration the general and ID specific orthopaedic literature (Kanis *et al.,* 2013, Kanis 2002, Angelopoulou *et al.,* 1999, Geijer *et al.,* 2014, Gonzalez-Aguero *et al.,* 2011) risks for poor bone health associated with people with ID were identified along with the identified risks recognised by the World Health Organisation (WHO 1994, WHO 2003) and the associated risks noted by the International Osteoporosis Foundation (IOF), the researcher compiled a risk factor framework to guide and structure the analysis. These risks were categorised into non-modifiable and modifiable risks with the anatomical systems guiding subcategorisation see *figure 5.0.*

Body mass index (BMI)

People reported their height and weight and BMI was calculated based on weight in kilograms divided by height in meters squared.

Physical activity

People also reported their level of physical activity (PA) classified by mild, moderate and vigorous PA for 10-20 minutes; more than once a week, once a week, 2-3 times a month, or hardly ever or never. An overall PA variable was then calculated based on the reported amount of time engaging in PA according to the World Health Organisation (WHO 2010) and Irish recommended guidelines (HSE 2010) of 150 minutes per week of moderate to vigorous intensity PA.

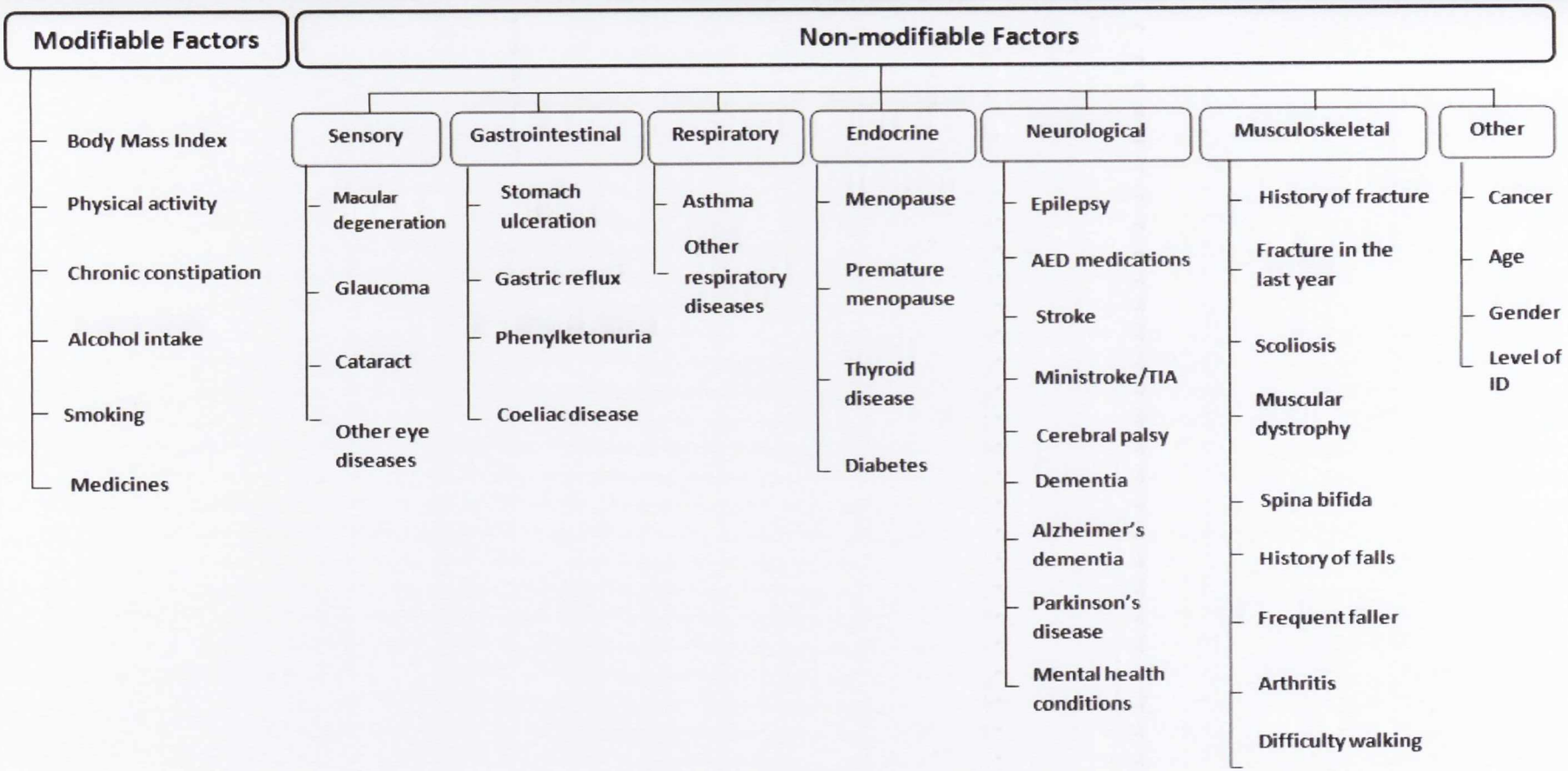


Figure 5.0: Non-modifiable and modifiable risk factors

5.4 Results

The prevalence of self-reported doctor's diagnosis of osteoporosis for this cohort is 8.1% (n=61). A binary logistic regression revealed those in the older age bracket (65+ years) were 6 time more likely to have a diagnosis than the younger cohort (40 – 49 years). Among the most prevalent risks identified were, female gender, having difficulty walking and having a diagnosis of arthritis. Reported rates of fractures (20%, n=152) were prevalent among this cohort, nevertheless rates for DXA screening were low at 16.8% (n=65) with just 11.1% (n=43) of those having had screening within the last 2 years.

5.4.1 Demographics

Table 5.1 presents a detailed description of the overall sample (N=753). The cohort comprised of 44.9% (n=338) men and 55.1% (n=415) women between the ages 40 and 90 years. Most people noted their level of ID at a moderate level however 58 participants failed to identify a level of ID. The majority of people lived in residential type setting (more than 10 people residing in one accommodation) with well over half, 69% (n=435), of the participants noting that they had some level of difficulty with verbal communication.

Table 5.1: Demographic profile of participants (N=753)

Variable		*n*	%	Totals
Gender	Male	338	44.9	753
	Female	415	55.1	
Age	40 – 49	275	36.5	753
	50 – 64	344	45.7	
	65+	134	17.8	
Level of ID	Mild	166	23.9	695∞
	Moderate	323	46.5	
	Severe/Profound	206	29.6	
Living circumstance	Indept./ Family	129	17.1	753
	Community group home	268	35.6	
	Residential setting	356	47.3	
Difficulty communicating	No difficulty	316	42.1	751≈
	Some difficulty	154	20.5	
	A lot of difficulty	104	13.8	
	Cannot do at all	177	23.6	
Bone Screening(DXA)	Within the last 2 years	43	11.1	389*
	Over 2 years ago	22	5.7	
Attended GP services		692	92.1	751
Attended geriatrician		19	2.5	
Medication use	Antiepileptic Drugs	287	38.2	
	Ca & Vitamin D supplementation	134	17.8	

∞Obs missing 58 (number of participants who did not identify their level of ID)
≈Two participants did not answer the question on communication
**This question was introduced post commencement of the overall study (n=477)*

5.4.2 Prevalence and identified risks for osteoporosis

In total 8.1% (n=61) of the participants identified that they had a doctor's diagnosis of osteoporosis. With regards to non-modifiable risks, fracture rates and falls were moderately high at 20.6% (n=152) and 26.7% (n=200) respectively, with a further 27% (n=214) reporting some level of mobility difficulties; 30.5% (n=225) reporting epilepsy with 38.2% (n=287) prescribed antiepilepsy medications (AEDs). In addition, 57.8% (n=238) of females reported having experienced the menopause with a mean age of onset for 98 participants at 47.02 years, (SD $\pm$5.63). Among the modifiable factors identified were low physical activity levels with over 70% not engaging in the recommended minutes[3] per week (WHO 2010), few people reported they currently or previously smoked (15.4%, n=116), with very low numbers of people reporting alcohol use at levels to incur bone interference (n=20). Levels of obesity were high with over 60.7% (n=358) reporting being overweight or obese with a small number of participants reporting underweight status (1.9%, n=6). Finally, 17.3% (n=130) reported chronic constipation (see **table 5.2** for details of all risks identified).

[3] The World Health Organisation recommends that adults aged 18–64 years and those 65 years and above should do at least 150 minutes of moderate-intensity aerobic physical activity throughout the week, or do at least 75 minutes of vigorous-intensity aerobic physical activity throughout the week, or an equivalent combination of moderate- and vigorous-intensity activity. 2. Aerobic activity should be performed in bouts of at least 10 minutes duration. 3. For additional health benefits, adults should increase their moderate-intensity aerobic physical activity to 300 minutes per week, or engage in 150 minutes of vigorous-intensity aerobic physical activity per week, or an equivalent combination of moderate- and vigorous-intensity activity.

Table 5.2: The prevalence of risk factors for poor bone health and increased fracture risk

Non Modifiable			*n*	%	Total
Musculoskeletal	Self-reported doctor's diagnosis of osteoporosis		61	8.1	752
	History of fracture		152	20.6	738
	History of falls		200	26.7	748
	Frequent faller		107	16.3	655
	Arthritis		81	10.8	749
	Mobility (walking 100 yds.)	No difficulty	537	71.5	751
		Some difficulty	82	10.9	751
		A lot of difficulty	52	6.9	751
		Cannot do at all	80	10.7	751
Neurological	Epilepsy		229	30.5	752
	Anticonvulsant Drug Use		287	38.2	752
	Cerebral palsy		43	5.7	752
Endocrine	Menopause	Experienced Menopause	238	57.8	412
	Mean age at menopause*	47.02 years, (SD ±5.63)			93
	Thyroid Disease		110	14.5	752
	Diabetes		61	8.2	747
Gastrointestinal	Gastroesophageal reflux		59	7.8	752
Sensory	All eye diseases~		21	27.1	749
Mental health condt.			355	47.5	747
Modifiable					
	Body Mass Index (BMI)	Healthy weight	217	37.2	584
		Overweight/Obese	358	60.7	584
	Chronic constipation		130	17.3	752
	Smoking	Current or past smoker	116	15.4	753
	Medicines	Hypnotics and Sedatives	100	13.3	752
		Antidepressants	204	27.1	752
		Anxiolytics	167	22.2	752
	Physical Activity	Meets RDA**	223	29.7	753
		Does not meet RDA**	528	70.3	

Only conditions with a prevalence above 5% are presented , those excluded were scoliosis, CVA, TIA, respiratory conditions, stomach ulceration, PKU, coeliac, all cancers, being underweight, history of alcohol >2 units 3-4 times a week and corticosteroids.

** 140 participants did not know their age at menopause*

~ All eye diseases include macular degeneration, cataracts, glaucoma and other eye conditions

***RDA=Recommended Daily Allowance*

5.4.3 Association of risks and self-reported doctor's diagnosis of osteoporosis

These risks were then examined in relation to those who had confirmed a self-reported doctor's diagnosis of osteoporosis (n=61). As outlined in **table 5.3** the most significant association with osteoporosis was older age ($p<0.0001$), female gender ($p<0.0001$), difficulty walking ($p<0.0001$) and a history of arthritis ($p<0.0001$). Strong statistical associations were also observed for those who had a diagnosis of epilepsy and were on AEDs ($p<0.0001$).

Table 5.3: Associations between self-reported doctor's diagnosis of osteoporosis and risk factors

		Doctor's Diagnoses of osteoporosis		
	Yes/No	%	n	p-value
Gender				<0.0001
Male		3.0	337	
Female		12.3	415	
Age				<0.0001
40 – 49		2.9	274	
50 – 64		9.0	344	
65+		16.4	134	
Level of ID				0.455
Mild		6.6	11	
Moderate		7.1	23	
Severe/Profound		9.7	20	
Living circumstance				0.156
Independent/Family		6.2	8	
Community group home		6.3	17	
Residential setting		10.1	36	
Type of ID – Down syndrome		9.5	14	0.455
Non Modifiable Risk factors				
History of fracture	Yes	13.2	151	0.008
	No	6.7	586	
Arthritis	Yes	22.2	81	<0.0001
	No	6.6	656	
Difficulty walking 100 yards	Yes	14.6	213	<0.0001
	No	5.6	537	
Epilepsy	Yes	13.5	229	<0.0001
	No	5.6	518	
Antiepileptic medications (AEDs)	Yes	11.8	287	0.003
	No	5.8	464	
Has gone through the menopause	Yes	15.1	238	0.016
	No	6.4	125	
Eye Disease	Yes	14.7	163	0.001
	No	6.3	584	
Modifiable risk factors				
Chronic constipation	Yes	15.4	130	0.001
	No	6.6	622	

Candidate variables that violated assumptions of chi squared test for associations due to expected cell count <5and are not included were memory impairment/Alzheimer's disease, diabetes, gastroesophageal reflux, smoking, hypnotics and sedatives, and antidepressants. All other candidate variables not presented did not achieve statistical significance

5.4.4 Binary Logistic Regression

A binary logistic regression was conducted to examine the influencing factors for self-reported doctor's diagnosis of osteoporosis, all odds ratios presented are crude odds ratios (OR). There is a definitive age gradient observed with those in the older age group almost 6 times more likely to have a diagnosis of osteoporosis than those in the younger age group (OR=6.53; 95% confidence interval (CI), (2.82-15.110). Females are over four times more likely to have a diagnosis (OR= 4.58; 95% CI 2.29-9.17) as were those with arthritis (OR= 4.07; 95% CI 2.21-7.48). Other significant factors can be observed in **table 5.4.**

Table 5.4 Binary logistic regression for osteoporosis diagnosis

Risk factors		Crude OR*	*P*-value[α]	CI~
Gender	Male	1	1	
	Female	4.582	**<0.0001**	2.289-9.172
Age	40 – 49	1	1	
	50 – 64	3.293	**0.003**	1.488-7.287
	65+	6.531	**<0.0001**	2.823-15.110
History of fracture		2.141	**0.009**	1.209-3.793
Arthritis		4.073	**<0.0001**	2.217-7.483
Epilepsy		2.547	**0.001**	1.501-4.320
Antiepileptic drugs (AEDs)		2.175	**0.004**	1.282-3.690
Difficulty walking		2.879	**<0.0001**	1.695-4.889
Experienced menopause		2.606	**0.019**	1.172-5.796
Eye disease		2.553	**0.001**	1.478-4.408
Chronic constipation		2.576	**0.001**	1.454-4.566

**OR=Crude Odds Ratio* *~CI= 95% Confidence Intervals*

[α]p-value set at 0.05

5.4.5 Consequences of osteoporosis

A foreseeable consequence of osteoporosis is fracture, an outline in **table 5.5** provides the profile of those who reported a history of fracture (20.6%, n=152). Proportionately, slightly more people with ID in the older age group (65 years+) reported having a history of fracture than those in the younger (40-49 yrs) or middle age group (50 – 64 years) at 22.6 % versus 17.8% & 21.8% respectively. Of those who reported fracture 12.5% (n=19) had DS with 8.9% fracture rate difference between those with and without DS (13.4% versus 22.3%). There was no gender difference with fractures reported at 20.7% in males versus 20.5% in females. The highest reported type of fracture was hip fracture at 12.6% (n=19), with no reports of vertebral fracture, similarly hip fracture was the highest reported type of fracture for those with DS (23%, n=5). Proportionately there were

slightly more fracture reports among those with mild level of ID and those living at home (22.0% and 24.2%) respectively.

Having epilepsy, being on an antiepileptic medicine, having difficulty walking and having a diagnosis of cerebral palsy were the most strongly associated factors with having experienced a previous fracture ($p < 0.0001$). In this study the reported prevalence for epilepsy was 30% (n=229) and of those, 4 in 10 reported a history of fracture, and the associated risk of epilepsy with osteoporosis was strongly significant ($p<0.0001$); reported AEDs medication usage was 38% (n=287) also significantly associated with fracture ($p<0.0001$). For those who had a diagnosis of epilepsy, 13.5% (n=31) reported a diagnosis of osteoporosis. As would be expected being a frequent faller or having a history of falling ($p <0.0001$) as well as osteoporosis ($p=0.008$) were also strongly associated with a history of fractures.

Table 5.5: History of fracture in association with risk factors

Variables	History of fracture	*n*	%	Number in sample	*p-value**
Gender					0.965
Male		8	2.7	329	
Female		84	20.5	409	
Age					0.395
40 – 49		47	17.8	264	
50 – 64		74	21.8	340	
65+		30	22.6	133	
Level of ID					0.830
Mild		36	22.0	164	
Moderate		63	19.9	317	
Severe/Profound		39	19.6	199	
Living circumstance					0.488
Independent/family		31	24.2	128	
CGH		50	19.0	263	
Residential setting		71	20.5	347	
Type of ID					**0.018**
Down syndrome		19	13.4	142	
Non-Down syndrome		133	22.3	596	
Other risk factors					
Doctors diagnosis of Osteoporosis	Yes	20	33.9	59	**0.008**
	No	131	19.3	678	
Epilepsy	Yes	64	28.4	225	**<0.0001**
	No	87	17.2	507	
AEDs	Yes	80	28.5	281	**<0.0001**
	No	72	15.8	456	
History of falls	Yes	65	43.0	197	**0.0001**
	No	86	16.0	537	
Frequent fallers	Yes	39	31.2	105	**<0.0001**
	No	86	16.0	537	
Cerebral Palsy	Yes	16	10.6	42	**0.004**
	No	135	19.4	695	
Difficulty Walking	Yes	67	31.8	211	**<0.0001**
	No	85	16.2	526	

p-value set at 0.005 statistically significance in bold*

5.4.6 Prevention and diagnosis

Almost one sixth of people (17.8%, n=136) were taking calcium/vitamin D supplementation, the majority were female (74.3%, n=101), were within the middle age bracket of 50 – 64 years (47.8%, n=65) and predominantly lived in a residential type setting (63.7%, n=86). For those who had a self-reported doctor's diagnosis of osteoporosis 70.5% (n=43) were prescribed calcium/vitamin D which means 3 in every ten with a diagnosis of osteoporosis were not prescribed these preventative measures. Of all those who had experienced a fracture over three quarters (75.7%) were not prescribed vitamin D, calcium or a combination. In total over 38% (n=284) did not drink milk, one of the best sources of dietary calcium.

The majority of participants (92.1%, n=692) reported they attended GP services in the last year however very few (2.5%, n=19) reported using specialist, like geriatrician services, the majority of whom were over the age of 65 years (n=12). Interestingly, 92.5% of the men and 86.0% of the women did not have a DXA within the last 2 years; similarly over 86% of those over 50 years had not attended for DXA and 81% of those over 65 years. The most statistically significant factor associated with non-attendance for DXA was difficulty mobilising (p=0.004). See **table 5.6** for profile of those prescribed vitamin D and, or calcium and factors associated with non-attendance for DXA screening.

Table 5.6: Factors associated with non-attendance for DXA and profile of those on vitamin D/calcium prescription

Variables		Did not attend for DXA* in the last 2 years					Prescribed Vitamin D/Calcium				
		Y/N	*n*	%	Number in sample	p-value	*n*	*Y/N*	%	No. in sample	*p-value*
Gender						0.043					0.0001
	Male		161	92.5	174		35		10.4	336	
	Female		185	86.0	215		101		24.2	417	
Age						0.010					<0.0001
	40 – 49		141	94.6	149		29		10.6	274	
	50 – 64		151	86.8	174		65		18.9	344	
	65+		54	81.8	66		42		31.3	134	
Level of ID						0.827					0.350
	Mild		90	88.2	102		25		15.1	166	
	Moderate		151	89.9	168		66		20.4	323	
	Severe/Profound		80	90.9	88		38		18.4	206	
Living circumstance						0.120					<0.0001
	Independent/family		84	94.4	89		9		7.0	129	
	CGH		136	88.9	153		40		15.2	264	
	Residential setting		126	85.7	14		86		24.2	356	
Other factors											
	Self-reported doctor's diagnosis of osteoporosis	Yes	11	39.3	28	0.0001	43	Yes	70.5	61	0.0001
		No	335	92.8	361		93	No	13.5	691	
	Difficulty walking	Yes	76	80.9	94	0.004	53	Yes	24.8	214	0.002
		No	269	91.5	269		82	No	15.3	537	
	AEDs	Yes	113	84.3	255	0.035	76	Yes	26.5	287	<0.0001
		No	233	91.4	134		60	No	12.9	465	
	History of fracture	Yes	64	83.1	77	0.073	37	Yes	24.3	152	0.026
		No	279	90.3	309		97	No	16.6	586	
	Epilepsy	Yes	88	84.6	104	0.100	71	Yes	31.0	229	<0.0001
		No	258	90.5	285		65	No	12.5	518	
	Menopause	Yes	97	82.9	117	0.103	76	Yes	31.9	238	<0.0001
		No	64	91.4	70		19	No	15.2	125	
	History of falls	Yes	86	91.5	94	0.362	49	Yes	24.5	200	0.007
		No	259	88.1	294		87	No	15.9	548	

**This question was introduced post commencement of survey*

5.5 Discussion

Despite advances in diagnosis and prevention of osteoporosis this study shows that people with ID present with many risks similar to the general population that increase the likelihood of reporting a diagnosis of osteoporosis. However for people with ID there are also additional risks such as epilepsy, AEDs prescription, early menopause among the females and genetic factors such as DS. Commonly identified risk factors observed among the general population such as smoking, alcohol, and corticosteroid use, are not consistently observed at rates to warrant concern.

In this study the high reported prevalence of epilepsy (30.5%) and AED medication use (38.2%) along with substantial levels of reported history of fracture (20.6%) is concerning.



Bone health is fundamental to ageing well and the maintenance of good bone health plays a vital role in achieving a healthy old age (WHO 2002). In this study just 8.1% presented with osteoporosis, however considering the insidious nature of the condition and the high worldwide prevalence (Kanis *et al.,* 2012, Odén *et al.,* 2015) the figure in this study is most likely an underestimation, especially considering estimated prevalence among people with ID varies from 17.1% (Zylstra *et al.,* 2008) to 43.9% (Bastiaanse *et al.,* 2014) in other reported studies. However caution must be exercised when considering these figures as prevalence can vary considerably depending on the methodology and definition applied. Nevertheless it is reported that people with ID present with increased risk of low bone mineral density (Aspray *et al.,* 1998, Center *et al.,* 1998, Baptista *et al.,* 2005, Jaffe *et al.,* 2005) and that osteoporosis continues to be under diagnosed and undertreated in the older population (Jasien *et al.,* 2012) which supports the results of this study. This is not surprising then considering that Srikanth *et al.,* (2011) report that the one area often neglected in physical health exams for people with ID is bone health status.

Worldwide osteoporosis is of concern especially for females (Compston *et al.,* 2013). Meunier *et al.,* (1999) recommend assessment of all females on cessation of menstruation. Over 57% of the females had experienced the menopause by an average age of 47 years, lower than the 52 years reported by the Health Service Executive (2008) and women in this study were found to be four times more likely to have a diagnosis, yet 86% of the females were not screened for osteoporosis. Such lack of investigation exposes the women with ID to an increased likelihood of experiencing fracture, an absolutely

preventable consequence of osteoporosis, potentially leading to devastating consequences for quality of life, independence, and physical and psychological well-being. However, in saying that, assessment can be fraught with difficulties for people with ID (McCallion and McCarron 2004). From fear of unfamiliar procedures, difficulty understanding complex test such as DXA, inaccessibility of equipment and environments, to difficulty in expressing their own health needs, all hinder or prevent people with ID engaging in assessment. These difficulties may form the basis of why it is noted that overall people with ID are less likely to have routine screening (Michael 2008) despite experiencing poorer health. Non-attendance at DXA screening is very evident in this study regardless of age or level of ID. Perhaps challenges experienced account for the low attendance especially considering that having a mobility difficulty was the most statistically significant factor (p=0.004) for not attending for DXA. Lying in a specific position for a certain length of time, poses challenges, along with following precise instruction. This requires a level of understanding that may be all too difficult for some people with ID. Therefore what is deemed as the gold standard for diagnosis (DXA) may not be suitable for people with ID. Perhaps these gold standards need to be rethought for people with ID and alternatives, such as the QUS, investigated and employed. Those who lived at home were also more likely not to engage in DXA screening; however this could possibly be because those at home were generally younger. Overall, levels of DXA scans were low, a finding similar to Dreyfus and colleagues (2014) who suggest the need for service provider awareness. Screening recommendations vary considerably and there are no specific recommendations for people with ID in Ireland. Due to policy change (HSE 2011) large residential type living and service delivery is changing and people with ID are moving to integrated community living. Here they are accessing community primary healthcare professional who may be delivering care to people with ID for the first time. Considering that some people with ID require a proxy to identify health care needs, educating physicians and caregivers is critical in light of the increased risk of osteoporosis in this population.

People with ID very often have more complex health needs and frequently present with older age conditions at a much younger age. These presentations could benefit from comprehensive assessment by specialist geriatric services that are skilled in the multidimensional diagnostic process which contributes to the development of co-ordinated and integrated plans of care. However, these services are commonly

chronologically determined, with those aged 65 years and over only having access. This immediately excludes those below this age despite presenting with older age conditions and may account for the small numbers in this study (2.5%) accessing such specialist services. This is unfortunate as if these services were expanded to include younger age groups of people with ID, accessing such co-ordinated multidisciplinary assessment earlier could facilitate earlier identifications of potential risk of older age conditions and comprehensive care pathways instigated.

The deterioration of the bone tissue associated with osteoporosis is reported to lead to increased bone fragility and risk of fracture particularly of the hip, vertebrae and wrist (International Osteoporosis Foundation [IOF] 2012). Hip fracture rates are reported just over three times higher at 12.9% than those reported by the general population study TILDA at 3.6% (Cronin *et al.,* 2011). Hip fractures can have devastating consequences on quality of life, independence and deplete an already compromised functional ability which is the case for many people with ID. Often the strict rehabilitation regimes are difficult for people with ID to follow and compliance becomes an issue, resulting in further loss of independence. The rates of fracture among those with DS is also substantially higher than those of the general population highlighting the need for robust assessment of those with DS which Angelopoulou *et al.,* (1999) notes as an independent risk for osteoporosis. It would be prudent for health care professionals to consider history of fracture when planning fracture prevention strategies in light of the potential of an increased likelihood of future fractures where history of fracture exists (Holloway *et al.,* 2015). One of the most intriguing findings in this study is the fact that there is very little difference in the fracture rates between males and females. Previous studies (da Silva *et al.,* 2010, Baptista *et al.,* 2005) have also identified high prevalence of low bone density among males especially those with DS and the findings here further confirm the misconceptions, that there is greater prevalence in females, which necessitates further investigations.

In this study the high reported prevalence of epilepsy (30.5%) and AED medication use (38.2%) along with substantial levels of reported history of fracture is concerning considering that the reported levels of osteoporosis are relatively low. It is known that AED medication interferes with bone metabolism thus compromising bone health. It is commonly assumed that fracture rates increase due to seizural activity, which does expose the person to increased falls risk (Sato *et al.,* 2001), however fracture rates have been shown to increase independent of seizure activity (Mattson and Gilda 2004). It

is quite probable that in this study osteoporosis is underdiagnosed and the bone compromise is increased in the presence of AED medication leading to increased fracture risk.

Bone is living tissue that requires stimulation to promote bone turnover and resorption making physical activity (PA) an important factor to prompt this process. In this study for 70.3% (n=528) of people PA levels were not at a level sufficient to accrue health benefit. Moreover, ambulation has been reported to play a significant role in helping limit the development of osteoporosis among people with ID (Tyler *et al.,* 2000, Zylstra *et al.,* 2008) and in this study those reporting difficulty with walking were 3 times more likely to self-report a doctor's diagnosis of osteoporosis (OR 2.9, $p<0.0001$, CI 1.695 – 4.889). Of those with mobility difficulty almost 1 in 5 ($p<0.0001$) reported a diagnosis of osteoporosis and 3 in 10 reported a history of fracture ($p<0.0001$). Engaging in PA is challenging for the best of us, it requires motivation and encouragement to participate and commitment to stay engaged sufficiently as to make it part of one's lifestyle and promote change. Those who can engage in PA ought to be encouraged to do so which requires effort and education for service providers, carers and the person themselves.

Difficulty with mobilising presents challenges for a substantial cohort in this study (28.5%, n=214). People with difficulty or who are not able to mobilise at all are dependent on secondary assistance. Educational programmes for carers and alternative activities to promote muscle strength and resistance that will stimulate bone turnover ought to be devised. Considering the low levels of exercise identified among this cohort and the positive consequences of introducing or increasing exercise regimes in people's lives it is recommended that consideration be given to improving lifestyle behaviours such as the introduction of physical activity programmes. These would also have the additional benefit of improving the overall muscular status of the person, improving balance and contributing to a reduction in the potential for falls.

It is interesting that those who reported eye disease were over two and half times more likely to report osteoporosis (OR 2.553, $p=0.001$, CI 1.478-4.408). Eye disease has no direct correlation with osteoporosis however is associated with falls risk, and is highly prevalent among older adults with ID, in fact previously McCarron *et al.,* (2013a) established a 71% prevalence of multimorbidity with eye disease identified as one of the highest occurring conditions. Therefore physical activity could also potentially contribute benefit to those who present with multimorbidity who may possibly be frailer. Another

emerging condition, chronic constipation, was also significant in the presence of osteoporosis. A possible explanation is perhaps that this is reflective of an imbalanced diet and may be reflective of inequity in the overall diet of the participant, chronic constipation could possibly effect vitamin and mineral absorption as well as having an effect on appetite further contributing to dietary imbalance and if left long term effect bone health.

Another contributing factor to poor bone health identified in this study are the low levels of vitamin D supplementation. Ireland is a northern hemisphere country and as such does not experience sufficient sunlight to promote adequate natural vitamin D synthesis; therefore the Food Safety Authority of Ireland has recommended supplementation for all citizens. However it was evident in this study that very few people (17.8%, n=136) were on vitamin D supplementation. It is concerning that 3 in every ten people with a diagnosis of osteoporosis were not prescribed this first line preventative and that over three quarters of those who had a history of fracture were not on vitamin D, calcium or a combination. It is known that having a history of fracture is a risk for future fracture and considering that over 38% did not drink milk, supplementation warrants follow-up investigation. By comparison, in the data reported by TILDA for the general population calcium and vitamin D was the most commonly reported food supplement, used by 31.4% of those who used food supplements (Peklar *et al.,* 2013).Vanlint and Nugent (2006) note vitamin D insufficiency is more prevalent among people with ID than the general population. However, Hirani and Primatesta (2005) report vitamin D deficiency at alarming levels among the general older population living in nursing homes. This is particularly concerning for people with ID who may have lived most or part of their lives in an institutional setting. Further the levels of AED use noted in this study are likely to have a negative impact on vitamin D levels. Despite the evidence of higher levels of supplementation among the TILDA participants their initial survey did not include people living in nursing homes (Barrett *et al.,* 2011).

For people with ID both receptive and expressive communication challenges pose difficulty (Kevan 2003). Over half of the cohort (58%, n=435) had difficulty communicating, with almost half of those (23.6%, n=177) reporting not being able to verbally communicate at all. McCarron *et al.,* (2011) noted that people with ID have some of their greatest challenges communicating with the health profession. McCarron *et al.,* (2011) also noted that people with ID too rarely receive easy-read information on their health or engage in health promotion contributing to reduced healthcare access for people

with ID, and health conditions going unrecognised and under treated (Bergström *et al.*, 2014, McCarron *et al.*, 2011, Kerr 2004). The vast majority of people with ID are dependent on others to manage their health need. If these carers themselves are unaware of the unique risks of osteoporosis very often barriers to healthcare are widened and inequalities increase (Cooper *et al.*, 2004).

This study supports further investigation of the bone health of people with ID and warrants systematic objective measurement of bone status using alternate methods focused specifically for people with ID.

5.6 Strengths and limitations

Undoubtedly the greatest strength of this study is the number of participants who contributed to the data. This study is based on the participants self-report of their doctor's diagnosis of osteoporosis from their clinical files and whilst opportunity was afforded to participants to access their medical files for accuracy no objective measures were conducted by the researcher. This study however confirms that the inclusion of objective measurement could contribute further to identifying specific risks among older adults with ID. Due to small numbers specific variables such as cerebral palsy and scoliosis, which in all likelihood contribute to the presence of osteoporosis, were excluded for some analysis. Finally the question on DXA screening was introduced post commencement of the study however on correlation analysis pairwise analysis command was utilised thus only completed cases are represented.

5.7 Conclusion

Despite high levels of risk, the reported level of self-reported doctor's diagnosis of osteoporosis was low and considering the insidious nature of osteoporosis one could only suspect that there is a hidden and undiagnosed level of osteoporosis among this cohort. The clinical outcome of poor bone health is fracture an event that results in unnecessary pain, physical disability and economic consequence impacting on people's daily living activities, quality of life and ultimately quality life years lived. The findings here support a need for robust risk assessment, regular checks and, for clinical practitioners to not only seek the obvious risk factors but also consider the specifics for ID. Further, although some risks are unalterable and inevitable such as AEDs required for epilepsy or the fact that the person has DS, there are amenable factors that require attention. Addressing inactivity,

Vitamin D supplementation, dietary balance and risks for falls recommendations may contribute to the amelioration of this silent and if left unhindered, debilitating disease. Considering the increased loading for people with DS it is interesting that they did not emerge at high risk. With this in mind the researcher extracted the data for those with DS in Wave 1 IDS-TILDA [2011] and examined their risk with regards diagnosis of osteoporosis. The next chapter will present the findings from that investigation.

CHAPTER 6

OSTEOPOROSIS AND PEOPLE WITH DOWN SYNDROME: A PRELIMINARY DESCRIPTIVE EXAMINATION OF IDS-TILDA WAVE 1 RESULTS

"Sometimes it's necessary to go a long distance out of the way in order to come back a short distance correctly"

- Edward Albee

Chapter summary

People with Down syndrome (DS) are a high 'at risk' group to develop osteoporosis. Increased morbidity associated with older age, higher prevalence of Alzheimer dementia, hypotonia, hypogonadism and thyroid disease are all features of DS and predispose this cohort to musculoskeletal complications. The Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing (IDS-TILDA) is a unique representative study of the ageing of adults with intellectual disability in Ireland. The sample (N=753) included 147 people with DS. Data was gathered on participants' health status, behavioural health, health screenings and activities of daily living. The prevalence of osteoporosis and related risk factors among people with DS was specifically examined. Of the 147 participants with DS, 9.4% reported a diagnosis of osteoporosis; a much lower figure than the prevalence of risk factors would suggest. Predisposing factors identified included higher than general population rates of thyroid disease (37.4%), epilepsy (19.3%), sedentary lifestyle (51.7%) and the majority of the females reporting having experienced menopause (61%). There was also little evidence of bone health population screening (8%) for people with DS despite the presence of such high levels of risk factors. Given the risk factor findings and the hidden nature of osteoporosis, underreported incidence among people with Down syndrome seems probable.

6.0 Introduction

Osteoporosis and osteopenia are chronic skeletal conditions characterized by low bone density and microarchitectural deterioration of bone tissue (WHO 1994, Kanis *et al.,* 1994, Kanis *et al.,* 2013). With ageing, enhanced resorption and decreased bone formation occurs leading to a net loss of bone mass and ultimately risk of osteoporosis and osteopenia (Datta *et al.,* 2008). Apart from net bone loss, the World Health Organisation (WHO), the International Osteoporosis Foundation (IOF) and bone health research literature, identify a number of non-modifiable and modifiable risk factors associated with the development of osteoporosis and osteopenia. Non-modifiable risks include female gender, increased age, family history of osteoporosis, being Caucasian and a history of prior fracture. Late menarche, early menopause and low endogenous estrogen levels are also associated with low bone mineral density (Kanis *et al.,* 2013, Kanis 2002, WHO 1994, WHO 2003). The modifiable risks include sedentary lifestyle, smoking, poor dietary habits, lack of sunlight and estrogen deficiency. Certain medications are also indicated as risk factors for example anticonvulsants and glucocorticoids (Beerhorst *et al.,* 2009). The higher the prevalence of risk factors has been shown to lead to a higher fracture rate with corresponding higher rates of morbidity and ultimately may have devastating consequences on the person's own independence, quality of life and overall health and well-being (Genant *et al.,* 1999, Kanis, 2002).

Osteoporosis and osteopenia have been widely investigated in the general population, however relatively under investigated among people with intellectual disability (ID), and especially among people with Down syndrome (DS) (Angelopoulou *et al.,*1999, Geijer *et al.,* 2014, Gonzalez-Aguero *et al.,* 2011, Guijarro *et al.,* 2008, McKelvey *et al.,* 2013). An increase in the older population of people with DS (Glasson *et al.,* 2002), also increases the probability of an increase in osteoporosis and osteopenia in this population.

6.1 Down syndrome, Ageing & Osteoporosis

Down syndrome (DS) is one of the most commonly known genetic causes of ID and its pathogenesis is well documented (Dierssen and DeLa Torre 2012). Specifically associated with DS are musculoskeletal disorders such as hypotonia, delay in motor skill development and joint hyperflexibility with risk of joint dislocation. It is noted that people with DS can have poorer health than the general population; however they are likely to have more

infrequent health checks or routine screening (Michael 2008). These inequities contribute to health problems going unrecognised, often compounded with the person's own inability to articulate their health needs. Srikanth *et al.,* (2011) report that one area often neglected in physical health checks for people with ID is bone health status.

Life expectancy among people with DS has extended however despite this mortality rates remain higher among people with ID than people in the general population. For example McCarron *et al.,* (2015) report a 19 year difference between people with ID in Ireland, which included people with DS, and the general population. For people with DS there are also issues of premature symptoms of ageing with related health decline and underlying musculoskeletal compromise which may predispose this group to osteoporosis and osteopenia (Bittles *et al.,* 2007, Glasson *et al.,* 2014, Roth *et al.,* 1996).

6.1.1 Non-modifiable risk factors affecting bone status among people with Down syndrome

Estrogen is required for adult bone maturation and maintenance both for females and males (Krassas and Papadopoulou 2001, Qiu *et al.,* 2013) and a greater lifetime exposure to estrogen reduces the risk of osteoporosis (Parker *et al.,* 2013). It has long been established that female gender related low estrogen levels are a risk factor for the development of osteopenia or osteoporosis as are late menarche and early menopause to which women with DS have been identified as being more prone (Schupf *et al.,* 1997). Schupf and colleagues report the median age of menopause for females with DS as 47.1 years. By comparison Gold *et al.,* (2013) report the median age of onset of menopause, depending on education, race and health status, between 52.17 years and 53.24 years among a cohort of females participating in the SWAN longitudinal study, a difference of between 5.07 – 6.14 years. It is also noted in the literature that hypogonadism, a feature of males with DS, is associated with reduction in pubertal growth spurt and thus they too because of low estrogen and testosterone levels (Seeman 1999), may be at increased risk for poor bone health.

Thyroid dysfunction is commonly identified among people with DS, with hypothyroidism the most common presentation (Karlsson *et al.,* 1998, Prasher 1999). Altered thyroid status impacts significantly on skeletal health. Karlsson *et al.,* (1998) reported that 35.3% of their cohort with DS had developed thyroid dysfunction with no gender difference, and with hypothyroidism most commonly occurring. Hypothyroidism

in youth is reported to result in delayed ossification and short stature whereas thyrotoxicosis in adulthood stimulates bone resorption and formation consequently increasing bone loss per remodelling cycle, in turn increasing the likelihood of later in life osteoporosis and subsequent fractures (Waung *et al.,* 2012).

Anticonvulsant medications (AEDs) contribute to disruption of bone formation and remodelling (Beerhorst *et al.,* 2009). As people with DS age the prevalence of Alzheimer's dementia and associated epilepsy increases (McCarron *et al.,* 2005) leading to an introduction of AED medications to control what can often present as refractory epilepsy. Beerhorst *et al.,* (2009) report on both first and second generation AEDs having a devastating effect on vitamin D and calcium metabolism, both essential elements of bone health, leading to increased risk of osteoporosis. The resulting poorer bone status, coupled with Alzheimer's disease and epilepsy, exposes the individual to an increased risk of fall and subsequent fracture, the devastating consequence of osteoporosis. However, Torr *et al.,* (2010) in their review warn that decline in adults with DS should not be assumed to be associated with Alzheimer's dementia as functional decline may be as a consequences of a number of underlying conditions especially sensory and musculoskeletal impairment. They note that preventative interventions are required to decrease premature mortality. Another contributing factor of note is the association of mental health conditions, such as depression, and Alzheimer's disease (Burt *et al.,* 1992, Collacott *et al.,* 1992). Here again it is the adverse impact of the associated medications that contribute to poor bone status. Anxiolytics, sedatives, and antidepressants primarily can effect postural balance leading to falls however some effect bone turnover and thus contribute to reducing bone mineral density (Vestergaard *et al.,* 2013).

People with DS have a predisposition to cervical neck abnormalities, however scoliosis or rheumatoid arthritis are not widely reported. Jaovisdha *et al.,* (1998) identified significant decrease in bone mass of the total hip and femoral neck among scoliotic women compared to nonscoliotic women and an incidence of 8.7% is reported among people with DS (Milbrandt and Johnson 2005). Rheumatoid arthritis (RA) is also associated with reduced bone mineral density. This autoimmune condition has been reported among younger cohorts with DS (Roizen *et al.,* 2014). Musculoskeletal conditions such as scoliosis and RA are prevalent risks associated with osteoporosis among the general population and could contribute to increased risk of osteoporosis among people with DS.

6.1.2 Modifiable risks of osteoporosis and osteopenia

The benefits of physical activity are widely investigated and reported among the general population (Penedo and Dahn 2005, Schwab and Scalapino 2011, Warburton *et al.,* 2006) with reports of resulting improved bone mass, stability, improved balance and overall quality of life (Carter *et al.,* 2001, Karinkanta *et al.,* 2007, Warburton *et al.,* 2001). Physical activity among people with DS is not as widely investigated with most reports focusing on Special Olympic participation or younger cohorts (see for example Balic *et al.,* 2000, Matute-Llorente *et al.,* 2013, Tsimaras *et al.,* 2003). However in their comparison of adolescents with and without DS, Matute-Llorente *et al.,* (2013) noted that those with DS who spend more accumulated time engaging in physical activity did gain health benefits, but none reached the minimum physical activity intensity or duration recommended to yield substantial health benefit. Physical activity training programme participation benefits for people with DS do occur (Tsimaras *et al.,* 2003), nonetheless Heller *et al.,* (2003) point out the influence of access barriers and carers' scepticism of value in outcomes. They highlight that when there is support for carers their perception of the benefits of exercise improves and they are more likely to support physical activity participation by persons with DS.

Among the general population it is noted that overweight and obesity has a cumulative effect on bone health, because adipose tissue retains vitamin D making it less available for bone metabolism (Wortsman *et al.,* 2000) and extra loading on joints and bones increases the likelihood of fracture if a fall occurs (Compston *et al.,* 2011). Although some adiposity is considered protective, obesity also increases the risk of type2 diabetes (Wentworth 2012). An unstable insulin state compromises the microarchitectural structure of the calcaneus increasing the risk of fragility fractures (Wongadee and Charoenphandhu 2015). The prevalence of overweight and obesity have been noted to be at higher levels among people with DS than the general population (see for example Rubin *et al.,* 1998, de Asua *et al.,* 2014) suggesting again that people with DS are at higher risk for bone health issues.

Vitamin D and calcium supplementation is purported to improve bone status with best results observed from those most compliant with medicine regime (Tang *et al.,* 2007). High levels of vitamin D deficiency partly due to low solar exposure have been reported by Frighi *et al.,* (2014) among those with ID and DS. Guijarro *et al.,* (2008) note that people

with DS had a lower volumetric bone mineral density associated with low levels of physical activity and low exposure to sunlight, necessary for the synthesis of vitamin D.

Known secondary causes of low bone mineral density include poor nutrition and gastrointestinal conditions such as coeliac disease, gastro-reflux disease, and chronic constipation, all highly prevalent among adults with ID and DS (Robertson *et al.,* 2000, Böhmer *et al.,* 2001). These conditions impact on dietary vitamin and mineral absorption resulting in an adverse impact on overall bone health.

6.1.3 Other contributing factors to poor bone health status

Insult to ocular functioning escalates the likelihood of an increase in postural sway increasing the vulnerability to falls (Lord *et al.,* 2010). Sensory impairment particularly cataracts, are highly prevalent among adults with DS (Van Allen *et al.,* 1999) and are associated with an increasing risk to falls.

People with DS have a higher level of health need and experience greater health inequalities by comparison to the general population (Cooper *et al.,* 2004). Health screening for this cohort occurs generally less and subsequently morbidities can go undetected (Piachaud *et al.,* 1998). There are several differing recommendations for bone density screening such as all women over the age of 65 years and all men over aged 75 years. Screening in people below these age groups is considered in the presence of major risk factors such as frequent or recent use of glucocorticoids (Compston *et al.,* 2009, Kanis *et al.,* 2008a, WHO 2003, WHO 2004). It is recommended that screening should be aimed at those at highest risk reducing the likelihood of treating healthy individuals. Given the risk loading for poor bone health in persons with DS screening for this population is warranted however Angelopoulou *et al.,* (1999) identified this area overall has been neglected.

The aim of this paper therefore is to examine the prevalence of risk factors predisposing people with DS to osteoporosis and osteopenia, and the relationship to a confirmed self-reported doctor's diagnosis.

6.2 Methods

Data pertaining to 147 people with DS participating in the IDS-TILDA study were utilised for this study.

6.2.1 Sample

In Ireland people with an ID who are in receipt of services are registered on a National Intellectual Disability Database (NIDD) which is managed by the Health Research Board (Kelly and O'Donohoe 2014). This database formed the sampling frame employed by the IDS-TILDA study. The inclusion criteria required people to be registered on the NIDD and be aged 40 years or older at the time of the study. IDS-TILDA recruited 753 people of all level of ID and from all living circumstances. A question on aetiology of intellectual disability was used to identify 147 persons with Down syndrome.

6.2.2 Ethics Review

Full ethical approval was granted by Ethics Committees of all the service providers involved in the study and also by the Faculty of Health Sciences Ethics Committee, Trinity College Dublin. Consistent with this review, all information was presented to participants in an easy-to-read format in order to obtain informed consent from the participants, and, family or guardians were also invited to support consent or provide agreement where applicable.

6.2.3 Measures

Following examination of the literature, the WHO guidelines (WHO 1994, WHO 2003, WHO 2004) and the International Osteoporosis Foundation (IOF) guidelines (Kanis *et al.*, 2013, Kanis *et al.*, 2009a, Crompton *et al.*, 2009, Kanis *et al.*, 2008a, Kanis *et al.*, 2007) common risk factors were identified and matched to the IDS-TILDA data collected in Wave 1 (see **table 6.1**).

Table 6.1: Measured Risk Factors for Osteopenia and osteoporosis

Known Risk Factors – IOF, WHO & Scientific Literature	IDS-TILDA Equivalent Data
Non-Modifiable	
Older age	Age
Female/ menopausal	Gender /Have you gone through the menopause
Hypogonadism	Have you any other chronic health conditions Male gender
Secondary osteoporosis	Doctors diagnosis of osteoporosis
Thyroid disease	Doctors diagnosis of thyroid disease
Other endocrine	Any other health conditions
Rheumatoid arthritis	Doctors diagnosis of rheumatoid arthritis
GIT conditions	Doctors diagnosis of gastrointestinal reflux
Coeliac Disease*	Doctors diagnosis of Coeliac Disease
Scoliosis*	Doctors diagnosis of Scoliosis
Diabetes Mellitus*	Doctors diagnosis of Diabetes Mellitus
Muscular dystrophy*	Doctors diagnosis of Muscular dystrophy
Cerebral Palsy*	Doctors diagnosis of Cerebral Palsy
Modifiable	
Sedentary lifestyle	Levels of physical activity Mobility – difficulty walking 100 yds.
Poor calcium intake	Dietary intake and supplementation
Vitamin D	Dietary intake, supplementation
Low body mass	BMI calculated as weight divided by height squared
GIT Condition	Doctors diagnosis of chronic constipation*
Anticonvulsant drug use	Anticonvulsant Medications
Corticosteroid drug use	Corticosteroid Medications
Anxiolytics, sedatives, neuroleptics and antidepressants	Anxiolytics, sedatives, neuroleptics and antidepressants Medications

**Conditions with low prevalence rates and subsequently not included in overall table stratified by age and gender*

Physical Activity

Ireland has adopted the WHO (2010) recommendations of at least 150 minutes of moderate intensity physical activity throughout the week with a focus on strength, balance and gait. Participants in this study were asked to describe their physical activity, both duration and intensity of engagement in terms of mild physical activity (minimal sweating or exertion for a period or 10 – 20 minutes such as bowls or golf), moderate physical activity lasting 10-20 minutes (light sweating or a moderate increase in breathing or heart rate, such as dancing or swimming) and vigorous physical activity lasting 10-20 minutes (causing heavy sweating and a notable increase in breathing or heart rate, such as vigorous swimming, running or jogging). They also reported frequency in terms of more than once a week, once a week, 1-3 times a month, or hardly ever or never. Responses were then

grouped into three categories (see **table 6.2**). People also identified if they had mobility difficulty by answering a global question on level of difficulty experienced when walking 100 yards.

Table 6.2: Physical activity categorisation

Sedentary	engaged in 10 – 20 minutes of mild, moderate or vigorous exercise less than once a week, 1 – 3 times a month or hardly ever or never;
Semi-active	engaged in 10 – 20 minutes of moderate exercise once a week and/or vigorous exercise 1 – 3 times a month
Active	engaged in 10 – 20 minutes of moderate or vigorous exercise more than once a week

Dietary Intake

Ireland uses a food pyramid of the main food groups (Health Service Executive & Department of Health and Children, (2012) reflecting the staple Irish diet. Portion sizes were illustrated for participants. For fruit, vegetables, potatoes, bread and cereal, meat, fish, poultry, milk cheese and yogurt, savoury and sweet snacks, fizzy drinks, water, tea, coffee and cordial people indicated the number of portions by frequency per food item i.e. more than four portions per day, 2-3 portions per day, 1 portion per day, 5-6 portions per week, 2-3 portions per week, 1 portion per week, 1-3 portions per month or never consuming this item. Each food item was then grouped and categorised in terms of being below, meeting or exceeding recommended daily allowances (Food Safety Authority 2011) taking into consideration age, gender and activity level.

6.2.4 Data Collection

Data was collected using a two phased approach of a pre-interview questionnaire, which was posted to the participant once consent had been received, and then the main questionnaire was administered via a face-to-face interview. On receipt of consent the field researcher contacted the participant and arranged an appointment for a time and place suitable to the participant. At this point the field researcher forwarded a pre-interview questionnaire which collected demographics, health conditions, healthcare utilisation and prescribed medications information. The main interview was conducted as a computer assisted personal interview. The main questionnaire consisted of 17 modules encompassing social, community, physical health, mental health and cognitive assessment (McCarron *et al.*, 2011).

6.2.5 Data Analysis

For the purposes of this study the Statistical Package for Social Science (SPSS) v22 was utilised for all analysis (IBM 2013). Preliminary analyses were initially carried out with included frequencies, distribution and identification of missing values. With consideration to missing data, valid percentages only are presented throughout the paper. Descriptive statistics were used to analyse the demographic data and overall relationships between variables were explored and stratified by age and gender.

6.3 Results

As may be seen in **table 6.3**, there were more females than males at 56% (n=83) vs 44% (n=62) respectively. Their ages were predominantly below 65 years, with 3 participants aged >64 years, all of whom were female. Participants lived in a variety of settings, community group home being the largest (42.9%, n=63) followed by residential type setting (37.4%, n=55). The majority of participants (70%, n=95) were within the mild to moderate range of ID. A large number of the cohort had no formal education (28.6%, n=42) with just 39.5% (n=58) reporting they attained some primary education. Whilst the vast majority visited their GP regularly very few used specialist services such as endocrinology (2.7%) or geriatrics (1.4%). Just 9.7% (n=14) of the participants reported a diagnosis of osteoporosis, all of whom were female, with the majority within the 50-64 year age category. Approximately 13% (n=19) reported having had a fracture, with prevalence twice as high among women (68.4%, n=14) compared to men (31.6%, n=5). Hip fractures were the highest reported type of fracture (26.5%, n=5) experienced. Finally only 17% (n=25) of all the participants reported taking vitamin D supplementation.

Table 6.3: Demographics of the Participants with Down syndrome (N=147)

Variables		n	(%)
Gender	Male	65	(44.2)
	Female	82	(55.8)
Age	40 – 49 years	79	(53.7)
	50 – 64 years	65	(44.2)
	65 + years	3	(2.0)
Living Circumstances	Independent/Semi	29	(19.7)
	Community Group Home	63	(42.9)
	Residential	55	(37.4)
Level of ID	Mild	20	(14.7)
	Moderate	75	(55.1)
	Severe/Profound	41	(30.1)
Education Achieved	No formal education	42	(28.6)
	Primary / some primary level	58	(39.5)
	Secondary level	1	(0.7)
	Other	18	(17.0)
	Don't know	21	(14.2)
Health Care Utilisation (within the last year)	GP	130	(88.4)
	Dental	85	(57.8)
	Optician	63	(42.9)
	Physiotherapy	34	(23.1)
	Dietician	31	(21.1)
	Hearing	25	(17.0)
	Endocrinologist	4	(2.7)
	Geriatrician	2	(1.4)
	Vitamin D supplementation	25	(17.0)

** Not all participants answered all questions*

6.3.1 Examining the risk factors for poor bone health.

6.3.1.2 Non-modifiable risks

The highest reported health condition was thyroid disease (37.4%, n=55) and again this condition was more predominant among females with the greatest prevalence among 50 – 64 year olds with only 2.7% (n=4) reporting having attended endocrinological services and 1.4% (n=2) gerontology services. Seizure activity was evident (19.3%, n=28), the most common type experienced was tonic clonic seizures (51.7%, n=15) and the majority of those experiencing epilepsy had attended an epilepsy clinic for review within the last year (75.0%, n=21). More females than males reported epilepsy with their rates increasing with age but the reverse were true for males, with a greater prevalence among the younger age category 40 – 49 year olds. As expected anticonvulsant use (20.4%, n=30) was reflective of the levels of epilepsy.

Among the female participants over 60% had experienced the menopause with the median age of onset 45.0 years (mean age of onset 44.8 years SD ±4.27) range 38 – 53 years respectively. A further 21% (n=31) were unsure if they had experienced the menopause.

Over 25% (n=40) of participants reported some level of difficulty with walking 100 yards of which over 10% (n=16) could not walk this distance at all. Middle aged women (50 – 64 years) presented with the greatest amount of difficulty (46.4%, n=19). For all non-modifiable risk factors see **table 6.4**.

Table 6.4: Non modifiable and modifiable risk factors associated with low bone mineral density according to gender and age

Variable		Total N=147		Women n=83			Men n=64		
				40-49 years n=39	50-64 years n=40	65+ years n=3	40-49 years n=40	50-64 years n=26	65+ years n=0
		n %		*n* %	*n* %	*n* %	*n* %	*n* %	*n* %
Non Modifiable									
History of Osteoporosis		14	(9.7)	3 (7.7)	11 (27.5)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)
Thyroid Disease		55	(37.4)	13 (33.3)	24 (60.0)	0 (0.0)	11 (27.5)	6 (25.9)	0 (0.0)
Epilepsy		28	(19.3)	3 (7.7)	13 (32.5)	1 (33.3)	7 (17.5)	4 (16.0)	0 (0.0)
Anticonvulsant Drug Use		30	(20.4)	4 (10.3)	12 (30.0)	1 (33.3)	7 (17.5)	6 (24.0)	0 (0.0)
Arthritis		21	(14.3)	6 (15.4)	10 (25.0)	0 (0.0)	4 (10.0)	1 (4.0)	0 (0.0)
History of fracture		19	(13.4)	6 (15.8)	8 (20.0)	0 (0.0)	2 (5.3)	3 (13.0)	0 (0.0)
Experienced Menopause		50	(61.0)	15 (39.5)	33 (80.5)	2 (66.7)	- -	- -	- -
	Don't know	13	(15.9)	5 (13.2)	7 (17.1)	1 (33.3)	- -	- -	- -
Mobility (walking 100 yds.)	No difficulty	107	(72.8)	32 (82.1)	22 (53.7)	1 (33.3)	34 (85.0)	18 (75.0)	0 (0.0)
	Some difficulty	17	(11.6)	4 (10.3)	9 (22.0)	0 (0.0)	3 (7.5)	1 (4.2)	0 (0.0)
	A lot of difficulty	7	(4.8)	0 (0.0)	3 (7.3)	0 (0.0)	2 (5.0)	2 (8.3)	0 (0.0)
	Cannot do at all	16	(10.9)	3 (7.3)	7 (17.1)	2 (66.7)	1 (2.5)	3 (12.5)	0 (0.0)
Modifiable*									
Body Mass Index (BMI)	Underweight	2	(1.7)	1 (3.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (5.0)	0 (0.0)
	Healthy weight	34	(29.6)	5 (15.2)	11 (42.3)	0 (0.0)	12 (34.3)	6 (30.0)	0 (0.0)
	Overweight	31	(27.0)	11 (33.3)	3 (11.5)	1 (100.0)	8 (22.9)	8 (40.0)	0 (0.0)
	Obese	48	(41.7)	16 (48.5)	12 (46.2)	0 (0.0)	15 (42.9)	5 (25.0)	0 (0.0)
Chronic constipation		10	(6.8)	0 (0.0)	6 (15.0)	0 (0.0)	3 (7.5)	1 (4.0)	0 (0.0)
Gastroesophageal Reflux		10	(6.8)	3 (7.7)	3 (7.5)	1 (33.3)	2 (5.0)	1 (4.0)	0 (0.0)
History of Smoking		7	(4.8)	1 (2.6)	2 (5.0)	0 (0.0)	3 (7.5)	1 (4.0)	0 (0.0)
History of alcohol intake		65	(44.2)	19 (48.7)	14 (35.0)	1 (33.3)	18 (45.0)	13 (52.0)	0 (0.0)
>2 drinks/day 3-4 days a week		2	(3.1)	0 (0.0)	0 (0.0)	0 (0.0)	1 (5.7)	1 (5.9)	0 (0.0)
Hypnotics and Sedatives		14	(9.5)	3 (7.6)	5 (12.5)	0 (0.0)	4 (10.0)	2 (8.0)	0 (0.0)
Antidepressants		26	(17.7)	3 (7.6)	10 (25.0)	0 (0.0)	9 (22.5)	4 (16.0)	0 (0.0)
Anxiolytics		17	(11.6)	4 (10.3)	4 (10.0)	0 (0.0)	4 (10.0)	5 (20.0)	0 (0.0)

**See below for other modifiable risk factors of dietary intake and physical activity*

6.3.1.2 Modifiable Conditions

In terms of BMI, the majority of participants were within the overweight and obese categories at 68.7% (n=79) with only 29.6% within the healthy weight category and just 2 participants identified as underweight. Almost 7% reported chronic constipation with a similar number reporting gastric reflux (6.8%, n=10). Few had a history of smoking (4.8%, n=7) with even fewer consuming more than 2 units of alcohol a day 3 or 4 times a week (3.1%, n=2). Reported levels of hypnotic & sedative, antidepressant and anxiolytic use were 9.5% (n=14), 17.7% (n=26) and 11.6% (n=17) respectively. No participants with DS were taking corticosteroids, a medicine noted by the WHO as among the higher contributor to risk of osteoporosis and subsequent fragility fracture risk (WHO 2003). See **table 6.4** for overall modifiable risks stratified by age and gender.

Dietary Behaviours

The majority of people ate three meals a day interspersed with snacks. All recommended daily allowances (RDA) are in accordance with the Scientific Recommendations for Healthy Eating Guidelines in Ireland (Food Safety Authority 2011). The most pertinent deficiency of dietary items linked to osteoporosis includes dietary calcium, vitamin D and intake of fizzy drinks particularly cola drinks. Many people with DS did not consume the recommended daily 5 portions a day of fruits and vegetables (87.7%, n=128), dairy (36.3%, n=53) and carbohydrate (23.8%, n=35). Considering gender, both men and women did not achieve daily recommendations for fruit and veg consumption, with men (93.8%, n=60) faring worst. With regards to sweet/fizzy drink/candy intake slightly over 50% met the suggested daily amount with the remaining 50% exceeding recommendations and men were more likely to exceed (59.7%, n=37). Considering fizzy drinks only, which includes cola, the majority 73.5% (n=108) had a low consumption (one or less 200 mls portion per week). Also, almost 43% (n=35) of women did not consume the recommended amounts of carbohydrates intake on a daily basis. Of concern in terms of bone health implications, over a third of both women and men (34.1% n=28 and 39.1%, n=25 respectively) did not attain the daily recommendations for dairy intake, a healthy source of calcium (RDA in Ireland is 1200 mg per day calcium) (see **table 6.5**).

Table 6.5: Food group intake recommendations according to gender distribution*

	Female (n=82)						Male (n=65)					
	Below Requirement		Meets Requirement		Exceeds Requirement		Below Requirement		Meets Requirement		Exceeds Requirement	
	n	%	*n*	%	*n*	%	*n*	%	*n*	%	*n*	%
Carbohydrates	35	42.7	38	46.3	9	11.0	-	-	65	100	-	-
Fruit and Veg	68	82.9	12	14.6	2	2.4	60	93.8	4	6.3	-	-
Dairy	28	34.1	46	56.1	8	9.8	25	39.1	36	56.3	3	4.7
Meat	2	2.4	78	95.1	2	2.4	4	6.3	60	93.8	-	-
Sweets	-	-	47	58.0	34	42.0	-	-	25	40.3	37	59.7

**Not all participants replied to all questions asked.*

Physical Activity

As outlined in **table 6.6** a large proportion of participants did not engage in physical activity at levels to accrue health benefit with 51% (n=76) of participants doing very little or no physical activity. This was particularly evident among the over 50 age group where just 38% (n=26) engaged in active or semi-active physical activity and even among the 40 – 49 year olds, levels of active engagement were very low with just 10.1% (n=8) reporting active levels of physical activity. Men led relatively inactive lifestyles with 54% (n=35) falling within the sedentary category and 40% (n=26) reaching only the semi-active level. Inactivity was also associated with level of ID with 83% of those with severe/profound ID being relatively inactive.

Table 6.6: Levels of physical activity engagement according to gender, age & level of ID

		Active		Semi-active		Sedentary		No. in sample
Age Groups		*n*	%	*n*	%	*n*	%	
Female (n=82)	40 – 49	5	12.8	20	51.3	14	35.9	39
	50 – 64	1	2.5	14	35.0	25	62.5	40
	65+	-	0.0	1	33.3	2	66.7	3
Male (n=65)	40 – 49	3	7.5	17	42.5	20	50.0	40
	50 – 64	1	4.0	9	36.0	15	60.0	25
	65+	-	0.0	-	0.0	-	0.0	-
Level of ID (n=136)*	Mild	1	5.0	14	70.0	5	25.0	20
	Moderate	8	10.7	34	45.3	33	44.0	75
	Severe/Profound	-	0.0	7	17.1	34	82.9	34

**Missing obs = 11 (People who did not identify their level of ID)*

6.3.1.3 Other factors impacting on bone health

Although numbers were small additional health conditions, as outlined in **table 6.7**, were reported by participants with DS that directly or indirectly affect bone health. These included coeliac disease at 5.5% (n=8), scoliosis (2.7%, n=4), diabetes (1.4%, n=2), muscular dystrophy (1.4%, n=2) and cerebral palsy (0.7%, n=1). Conditions indirectly affecting bone health, namely risk of fall and subsequent fracture, were notably higher, these included cataracts and Alzheimer's dementia at 29.9% (n=44) and 15.6% (n=23) respectively.

Table 6.7: Other health conditions directly and indirectly effecting bone health

Other health conditions directly effecting bone health	*n*	%
Coeliac disease	8	(5.5)
Scoliosis	4	(2.7)
Diabetes mellitus	2	(1.4)
Muscular dystrophy	2	(1.4)
Cerebral Palsy	1	(0.7)
Health conditions indirectly effecting bone health		
Cataracts	44	(29.9)
Alzheimer's dementia	23	(15.6)
Asthma	7	(4.8)
Cancer (all reported)	2	(1.4)

6.3.2 Reported Bone Density Screening

Only 7 people reported having had dual-energy x-ray absorptiometry (DXA) scan and just 3 people within the last 2 years. No men reported having had a scan no matter what age. Further, no person with a history of fracture reported having had a DXA and screening levels were low among those reporting risk factors such as experience of menopause (n=50), thyroid disease (n=55) and epilepsy (n=28) at 30% (n=6), 21.4% (n=6) and 30.8% (n=4) respectively. For the DXA question there were only 87 participant answers.

6.4 Discussion

Reported attendance at DXA screening was low (7/147) and no men reported having attended for screening.



Down syndrome itself has been shown to increase the risk of the presence of osteoporosis and osteopenia due to the number of risk factors associated with DS, such as hypotonia or hypogonadism (Angelopoulou *et al.,* 2000, Angelopoulou *et al.,* 1999, Sepúlveda *et al.,* 1995, Kao *et al.,* 1992). Findings here concur that people with DS are a high 'at risk' group. The confirmed prevalence of thyroid disease, early menopause and specific medications that interfere with bone metabolism, and the levels of inactivity and sedentary lifestyle, all culminate in increased likelihood of the development of osteoporosis and osteopenia.

Osteoporosis is receptive to treatment and preventable; however in this study active health promotion was not evident. For example, physical activity engagement at levels that would accrue health benefit was not evident. Although rates of physical activity were very low, some participation is better than none and promoting engagement in exercise that is fun, enjoyable and diverse needs to be encouraged. With imagination and resourcefulness people with DS can be encouraged to engage however, as Heller *et al.,* (2003) indicates, it is their carer's and supporters that make the difference and lead by example.

Attendance at specialist services such as endocrinology or gerontology was also low and although people attended their GP regularly the levels of vitamin D supplementation was low. In Ireland, a northern hemisphere country, it is acknowledged that sunlight levels are considered low and supplementation is recommended (Genant *et al.,* 1999). The Irish recommendations for Vitamin D supplementation is 5ugs for people up to 50 years and 10ug per day for those aged 51 years and over (Food Safety Authority 2011). Genant *et al.,* (1999) suggested that further pharmacological interventions ought to be targeted at those with highest risk of fracture but little evidence of this was found for people with DS. The finding that overall dietary balance in the sample was poor suggests future research ought to examine whether dietary status is an issue of poor choices or a lack of awareness among health professionals and carer's. Considering that many of the participants in the study lived in supported settings, where the daily dietary decision making may be taken out of their hands, these findings support the implementation of

education to improve overall diet and consideration for supplementation for people with DS.

Levels of diagnosed osteoporosis were not particularly high, but low screening rates with high presence of risk factors perhaps suggest hidden undiagnosed disease, especially among males. No male reported the presence of osteoporosis or osteopenia, nor having had a DXA, despite risk factors associated with males with DS for example hypogonadism. In addition, 13.4% of the participants in this study reported having experienced a fracture, with hip fractures occurring at twice the rate reported by those in the general population study The Irish Longitudinal Study on Ageing Wave 1 results (TILDA), (Barrett *et al.,* 2011); yet overall only 8% reported having had a DXA scan. Lack of clinical assessment for people with DS has previously been identified as an area of neglect in Ireland (Doyle *et al.,* 2011). Consideration ought to be given to the van Allen *et al.,* (1999) suggestion of regular systematic screening for people with DS especially as the WHO also recommends such screening for all individuals with one or more strong risk factors. They would argue menopause is one such factor, and as can be seen here a large number of the women had gone through menopause, and; perhaps hypogonadism among males would be another. Whilst they do concede that population screening is not a cost effective approach, making more widely available techniques such as quantitative ultrasound ought to be explored (Genant *et al.,* 1999). The TILDA study engaged in bone screening for general population participants, however confirmation of the self-reported doctor's diagnosis of osteoporosis was much less than numbers found to meet criteria through screening. In the TILDA study they found that 86% (n=1365) with objectively measured evidence of osteopenia and 72% (n=218) osteoporosis did not have a doctor's confirmation of diagnosis (Cronin *et al.,* 2011). Overall, as Haveman *et al.,* (2010) would purport, greater surveillance of health risks is seen as a way to improve the health status of older adults with ID.

It must be highlighted that people with DS do experience communication challenges and expressing their health needs can often pose challenges. However, as people with DS age their increased vulnerability to the development of Alzheimer's dementia is well recognized and perhaps a contributing factor to other clinical signs and symptoms of underlying medical conditions going unrecognised (McCarron and Lawlor 2003). Alzheimer's dementia also increases the person's susceptibility to falling thus increasing the risk of fracture. Instigating measures to prevent and address poor bone

health at an early stage would be preferable, along with increasing carer's awareness of the potential and insidious nature of osteoporosis which can lead to fracture, and possible further damaging consequences.

6.5 Limitations

Limitations to this paper must be recognised; the group studied is larger than most published papers on DS and were drawn from a representative sample of people with ID but there were small numbers in some of the subgroups considered. Further, the presence of osteoporosis was based on a doctor's reported diagnosis and was not objectively measured.

6.6 Conclusion

Evidence emerges from this study that identified risks which include thyroid disease, epilepsy and its associated medications, inactivity and dietary imbalance are at levels which, based on prior general and DS population studies, would increase the risk of developing osteoporosis and osteopenia. Coupled with the predisposition that already exists among people with DS this is a recipe for future deleterious consequences.

Given the increased risk loading for osteopenia and osteoporosis and the low levels of screening among those with DS, it is questionable that the reported low prevalence of osteoporosis is reflective of the status of bone health among this population. Therefore further investigation utilising objective measurement of bone health is warranted.

In recognition of this, objective measurement of bone health was introduced in Wave 2 [2014] of the IDS-TILDA study. The next chapter will present the design and development of the objective measurement including the underpinning philosophy to promote inclusion and engagement of those involved in the objective measurements.

CHAPTER 7

DESIGN AND DEVELOPMENT OF THE HEALTH ASSESSMENTS FOR IDS-TILDA WAVE 2

"Getting told I can't do something gives me all the motivation I need to get things done."

- Kushandwizdom

Chapter summary

The Intellectual Disability Supplement to the Irish Longitudinal Study on Ageing (IDS-TILDA) is designed to capture all domains of health and wellbeing as one ages, and is directly comparable to *The Irish Longitudinal Study on Ageing* (TILDA) investigating ageing among the general population in Ireland. However, unlike TILDA in Wave 1 IDS-TILDA did not include objective health measurements. This was partly due to the cumbersome nature of the ethics procedures at the time and also on the advice of the Scientific Advisory Committee who felt that this element could be delayed until a future wave. Therefore the objective measures were not introduced until Wave 2. Driven by the success of the first wave and by the increasing interest in the study it was felt that objective measurement at this time would be more favourable. However, evidence based guidelines on engaging persons with ID in health assessment and health monitoring is limited. To that end the researcher adopted an emancipatory philosophy to underpin the development of the objective measurement protocol. A suite of 8 health assessments were included in a process purposely named *'The Health Fair'*, to avoid negative connotation of being measured or tested. The process was designed to be mobile and overcome barriers such as communication challenges or access, that people with ID face. Following initial pilot testing all participants from the IDS-TILDA study were invited and the researcher travelled throughout the country engaging 604 participants in the *Health Fair.* Of those who attended the *Health Fair* females were significantly more likely than males as well as those with very good to excellent rated health. Potential inhibitors included anxiety about what was going to occur, behaviours that challenge, communication challenges and physical health or mobility issues. Over 85% engaged in the *Health Fair* which is reflective of the success of the process and the achievement of the participants. It is hoped that the protocol developed will contribute to guidelines for the health assessment of people with ID and in doing so contribute to developing a greater understanding of their health pathways to improve their health care delivery and services now and to the future.

7.0 Introduction

The World Health Organisation (WHO) predicts significant growth in chronic disease among the older population by 2050 (World Health Organization 2015b) and this is likely to be even more marked in people with ID, given the increased risk loading for chronic disease as well as inequities in health care provision for this increasingly at risk population (Emerson and Baines 2011). Similar to current efforts in the general population, understanding the contributors and determinants of health and wellbeing in people with ID as they age is critical. While there has been an increase in ageing studies in the field of ID, there have been a number of methodological challenges including small sample sizes and different outcome measures, with few population based longitudinal studies, thus limiting the ability to compare with general ageing studies. In addition, many studies have been survey based with limited objective measures of physical health.

Health and healthcare inequalities among people with ID have been highlighted for many years (Krahn *et al.,* 2006, Alborz *et al.,* 2005, Kerr 2004, Kerr 1998). Inappropriateness or lack of available health care information, fragmentation and discontinuity of service provision especially for those with multiple and complex needs, and lack of empirical evidence on health determinants to underpin healthcare policy (Tracy and McDonald 2015, Emerson and Hatton 2013, Alborz *et al.,* 2005, Sowney and Barr 2004), are among some of the challenges people with ID face. These barriers increase the person's exclusion, impact on quality of life and contribute toward stigmatisation and disenfranchisement of people with ID. Inaccessible facilities, equipment and communication challenges continue to compromise the health care experience of many people with ID, and contribute to a lack in understanding of the health needs of people with ID.

Policy directions in Ireland and internationally (Health Service Executive 2011, von Bogdandy 2000) has focused on supporting older adults with ID to live in more community based settings and to access more generic health care services, however the conditions necessary to support equality of access is poorly understood and evidence based guidelines on engaging persons with ID in health assessment and health monitoring is limited. Preparing health care professionals including doctors and nurses, as well as all members of the multidisciplinary team, to support people engage in their health, monitoring and assessment is imperative.

7.1 Aims

The aim of this chapter therefore is to present the methods employed in designing and undertaking a suite of health assessments as part of Wave 2 of the IDS-TILDA study including the underpinning design philosophy. This design is offered to support researchers and practitioners in clinical practice improve assessment practices with people with ID.

7.2 Methods

7.2.1 Including Health Assessments

Integrating objective health measures into population studies promotes validation of self-reported health data and provides objective identification of risk factors of ill health. Objective measures can also promote more sophisticated analyses of disease pathways. As this paper is primarily related to the design and development of the objective health measures the reader is directed to McCarron *et al.*, (2011) and Carroll *et al.*, (2014) for a broad overview of the methodology of the IDS-TILDA study. Objective measures were new to the IDS-TILDA study and their incorporation in Wave 2 was influenced by results from similar studies (TILDA, ELSA) that suggested most participants found completing objective measures to be a positive experience. All equipment utilised in the IDS-TILDA health assessments replicated the equipment demonstrated valid, reliable and utilised in TILDA supporting direct comparison with this general population study. A fundamental difference was that the researcher who developed the protocol and administered the objective measures had extensive and considerable experience working with people with ID. This enabled key skills to be applied in developing a process suitable, for personnel working in the area of ID to complete, following sufficient training.

7.2.2 The objective health measures

Assessments included anthropometrics; height and weight (to calculate BMI), cardio metabolic measures; waist and hip measurement (waist to hip ratio calculated), blood pressure measurement (2 sitting and 2 standing to determine orthostatic hypotension), mobility, muscle strength and bone health measures; timed up and go test, grip strength (2 measurements on each hand), and quantitative ultrasound of the calcaneus (for bone

assessment). **Table 7.1** details the full list of the objective health measures and equipment employed.

Table 7.1: Objective health measures included in the IDS-TILDA study

Objective health measurement	Number of measurements	Equipment or test used
Height	1	Seca 217 portable stadiometer
Weight	1	Seca 803 digital scale
Waist size	2	Seca tape 201 Ergonomic
Hip size	2	Seca tape 201 Ergonomic
Blood pressure	4 (2 sitting and 2 standing)	Omron M10-IT Digital Automated Monitor
Grip strength	4 (2 on each hand)	Jamar Hydraulic Dynamometer
Timed up and go	1	Standard chair (45 cms), tape measure, high visibility tape and a stopwatch
Bone assessment	2 (both heels scanned)	GE Lunar Achilles Ultrasound

7.2.2.1 Anthropometrics

Height was measured using the mobile stadiometer seca 217 which is designed specifically with mobility in mind; it is easily assembled and has an adjustable wall spacer to ensure secure stability without any fitting requirements. For weight measurement the seca 803 digital portable flat scales was chosen. This had a large surface area and is almost flush to the floor, thus ensuring people do not have a large step up, which might pose difficulty. Body mass index, used for assessing degree of overweight as per WHO cut-off points was calculated as weight in kg divided by height in meter squared (kg/m^2) (World Health Organisation 1995). The mid upper arm circumference and ulna length measurement were offered and utilised for those who were immobile, afraid, found it difficult or refused to stand on the stadiometer or secca scale. This provided validated alternate measures of body mass index and height (BAPEN 2012).

7.2.2.2 Cardio metabolic measures

In the *Health Fair*, blood pressure was measured using a digital automated oscillometric blood pressure monitor (Omron M10-IT, Omron Inc.). In total four measures were taken, 2 sitting and 2 standing, with one minute rest between measures. Simultaneously, the Omron M10 recorded the pulse rate and any identified irregularities were noted and recorded.

Measuring body fat is expensive and complicated however waist circumference and waist to hip ratio has become accepted as surrogate measures of abdominal fat mass. A seca 201 ergonomic circumference measuring tape was used for waist and hip measurement. The waist circumference was measured at the level midway between the lower rib margin and the iliac crest, with the participant breathing out gently, standing erect with arms by their side wearing one layer of light clothing. The waist to hip ratio is calculated by dividing the waist measurement by the hip measurement. **Table 7.2** presents the WHO cut-off points stratified by gender for the increased risk of metabolic disease (World Health Organisation 2011).

Table 7.2: World Health Organization cut-off points and risk of metabolic complications

Indicator	Cut-off points		Risk of metabolic complications
	Male	Female	
Waist circumference	>94 cm	>80 cm	Increased
Waist circumference	>102 cm	>88 cm	Substantially increased
Waist–hip ratio	≥0.90 cm	≥85 cm	Substantially increased

Adapted from the WHO, 2011b

7.2.2.3 Mobility, muscle strength and bone health

The timed up and go (TUG) test was chosen to replicate the general population study TILDA and also to assess the functional mobility of participants. TUG assesses proximal muscle strength, balance and executive function (Podsiadlo and Richardson 1991). The participants were instructed to get up from a sitting position, walk at their usual pace, a distance of 3 meters (which was indicated with bright coloured tape measured from the front leg of the chair to the point on the floor), turn around at this point, walk back to the chair and sit down. A standard chair measuring 45cms from the floor to the top of the arm rest was used throughout data collection. As an extra precaution the chair was always set up against a wall to provide secure support. The procedure was timed in seconds with a seca stopwatch and recorded. The researcher instructed the participant and demonstrated what was required. Before the timed assessment the participant was offered a practice run if they wished.

A Jamar hydraulic dynamometer was used to assess grip strength. This has been found to be a valid and reliable instrument for measuring hand strength (Mathiowetz *et al.*,

1985). The peak-hold needle automatically retains the highest reading until reset and it has easily adjustable anatomical grip positions. Dominance is a difficult concept to establish with people with ID as they may not understand what is or know their dominant hand. This was assessed using three pre-assessment skills. Initially the person was asked to sign their consent and the researcher noted the hand used for signing. Then the person was asked which was their dominant hand (strongest or which one they used for doing most things). If there remained a difficulty the researcher got the participant to squeeze two of her fingers on each hand and noted which side was stronger. Four grip strength readings were obtained, 2 from the right hand and 2 from the left, all results were recorded. To avoid the introduction of parallax error a particular method of reading the result was included in health assessment protocol (see appendix 8).

The GE Lunar Achilles (QUS) provides a reliable and convenient method for the evaluation of osteoporosis and fracture risk. It is a lightweight portable machine previously used in large-scale screening studies in the general population and for people with ID. It has also been successful in identifying people with osteoporosis and at risk of fracture (Mergler *et al.,* 2010). The International Society of Clinical Densitometry (ISCD) official position is that the QUS is justified as a tool of measurement in the absence of DXA, however, note that only validated devices scanning at the os calcis (heel) are recommended (Krieg *et al.,* 2008). The device utilised in this study met these criteria. The device calculates three parameters, broadband ultrasonic attenuation (BUA in dB/MHz), Speed of Sound (SOS in M/s) and Stiffness Index (SI, %) which it then converts into those at high risk of fracture (osteoporosis), those at moderate risk (osteopenia) and those at low risk (normal). These parameters are and expressed as QUS t-scores by the device. These parameters have been translated into risk estimate thresholds for clinical application (Hans *et al.,* 2003). In an effort to minimize the inappropriate use of QUS, Hans and colleagues use 90% sensitivity to define low fracture and osteoporotic risk and a specificity of 80% to define persons as being at high risk of fracture or of being osteoporotic. They combined their fracture model with their hip DXA osteoporotic model and found t-score parameters of -1.2 and -2.5 for the QUS Achilles stiffness index to determine those; a low risk (normal) above -1.2; moderate risk (-1.2 to -2.49; osteopenia); and $\geq$ -2.5 at high risk osteoporosis. These parameters have been utilised in this study. Two measures were taken, one left and one right, any abnormalities were noted. For those who were immobile but amenable to

the assessment a raised block system was designed to elevate the QUS so the person could comfortably and accurately place their foot in the machine from their wheelchair.

7.3 The Design Philosophy

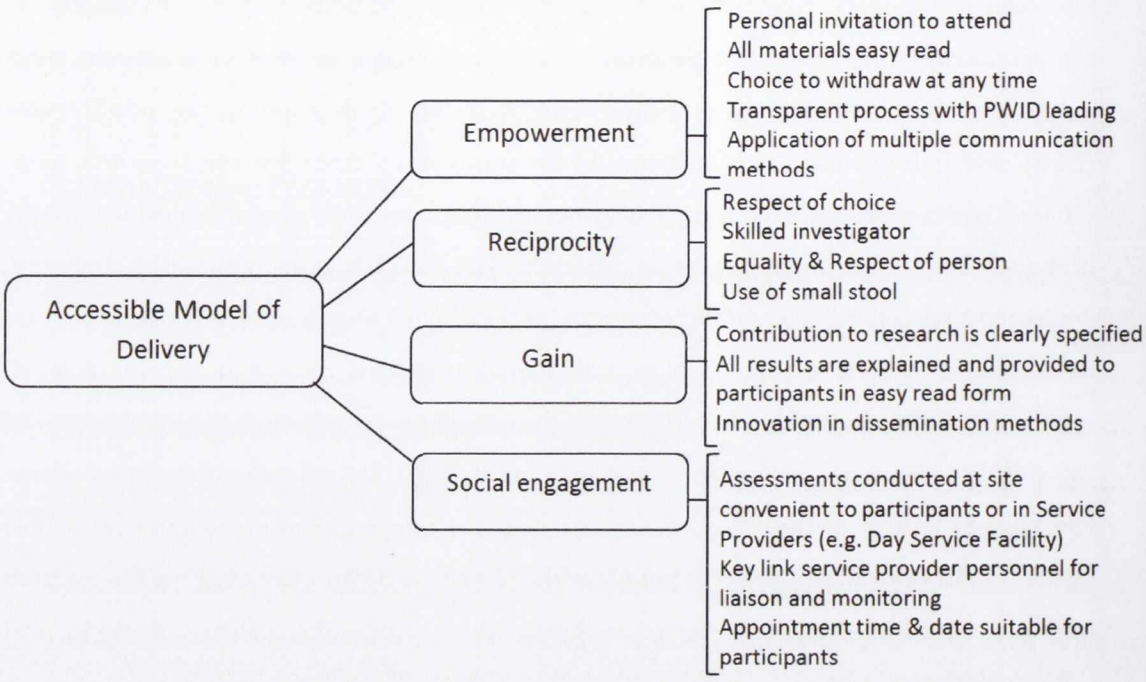
Designing a health assessment process to be included in any large scale research is a complex and challenging task and can be more challenging when participants have an intellectual disability. The health disparities literature is replete with examples of failed initiatives, imperfect understanding of how health care and health assessments may be successfully delivered and, paternalistic rather than inclusive intervention and research efforts (Taggart and Cousins 2014, Emerson and Baines 2011, Katterl and Bywood 2011, Chew *et al.,* 2009). The differences in health experiences, the challenges in communication and tolerance of assessment for people with ID require a different approach. To that end, the researcher adopted the key fundamentals of *Emancipatory Research Design Philosophy* (Oliver 1992), that is, social engagement, reciprocity, gain and empowerment to build an accessible model of health assessment delivery.

> *"Emancipatory research is about the demystification of the structures and processes which create disability, and the establishment of a workable dialogue between the research community and disabled people. To do this, researchers must put their knowledge and skills at the disposal of disabled people".* (Barnes 1992: 122)

Underpinning the approach to the health assessment design, accessible materials and health assessments, with this philosophy is a deliberate shift away from emphasising the individual's impairment onto the environment, society or other such barriers that enable disadvantage to continue (Barnes 2002). The social engagement element incorporates interaction and exchange between the researcher and participant. The focus is on the activity or shared task, integrating the perspectives of both parties working together as equals. Supporting this is the second element of emancipatory research, namely reciprocity, which represents a research environment of mutuality and respect contributing to both persons gaining an understanding of the value in the exchange that takes place. This blends almost translucently with the ethos of mutual benefit which refers to gain. The extent to which the research and researcher seeks to integrate the knowledge gained with interventions and, policy and social change, to improve the health and quality of life of the

research participants. The final and the most integral component of the philosophy i.e. empowerment, which is the promotion of the participants, endorses them as the most imperative factor in the process. Empowerment shares the decision-making of all phases of the research process and transforms the locus of control from the researcher to a more shared endeavour enabling the participants to articulate their views, contribute to the experience and be true partners in research. The emancipatory philosophy embodies reflexivity, whereby the researcher examines not only what they are doing but how they are doing what they are doing. Through the emancipatory lens the researcher questions all actions with the ultimate goal of improving the overall process through the involvement of all stakeholders vested in the research (Butts and Rich 2013). Adopting this approach was an attempt to ensure the '*rip off research tradition*' for inappropriate exploitation of people with ID did not occur; nor were people with ID excluded from any stage of the research process, thereby effectively disenfranchising them from the control, creation and dissemination of information about themselves (Oliver 2002, Barnes 2002).

These principles were then operationalized for the project (see *figure 7.0*) and the model for health assessment developed included participants contribution in protocol development, a multi-tiered quality review before going to the field, and the roll out of the health assessments with each of the principles embedded within. The first reflection of this design was that the research assessment process would be referred to as '*The Health Fair*'. This was to avoid negative connotations of being tested and promote a more enjoyable interactive experience.



*PWID – People with intellectual disability

Figure 7.0: Applying the emancipatory research design to the health assessment process

7.3.1 Building an accessible model of delivery to ensure reasonable adjustment

The accessible health assessment model considered (1) what the assessments should be called to reduce potential for negative connotations, (2) how the person to be included in the study was contacted, (3) where the assessments would take place, (4) the process of meeting and creating a welcoming, relaxing and respectful atmosphere, (5) the method of gaining consent, (6) the way the measures were administered, (7) the behaviour of the researcher to reduce overpowering or standing over the person, give clear instruction and demonstration, undertake assessments only when the person was happy to do so, (8)developing a shared understanding of the importance of the activity (9) adjustments of the research data collection methods. See *Health Fair Protocol* (appendix 8) for detail of each of these points. Attention to building collaboration with all stakeholders and clear consideration to the accessibility of materials promoted the realisation of the intended emancipatory design.

7.3.2 Working groups and collaboration

Seeking additional input reflected a belief that people with ID are best positioned to identify their own needs and experiences. Initially the researcher worked with an independent self-advocate to draft the explanatory materials and overall health assessment process including the accessible consent process, explanatory easy read material (see appendix 8) for the assessments and the plan for the overall roll out of the *Health Fair*. The researcher then organised focus group consultations with a number of independent self-advocacy groups throughout the country.

Three different advocacy groups reviewed and commented on the material and process making a number of suggestions and changes that were incorporated into assessment process. For example, the groups suggested the inclusion of processes to assist people with visual or hearing impairment, the researcher therefore included Lámh, modelling, hand-over-hand as well as visual demonstration. Suggestions around improving the picture size and quality were also helpful contributions along with the overall agreement that this would be an important and useful part of the study. Finally, an independent advocate reviewed the finalised material for user friendliness and ease of understanding. In tandem, the researcher consulted with the IDS-TILDA scientific advisory board that also guided and reviewed the overall health assessment, the accessible material and the final process of collection.

7.3.3 The development of the accessible material for the health assessments

It is encumbant on researchers in the field of ID to acquire the necessary skills to ensure that the views of people with ID, who may have verbal communication difficulties or use an alternate means of communication entirely, are understood and included. In keeping with the emancipatory design the researcher developed easy-to-read explanatory and supporting material that were included in the invitation pack, an easy-to-read letter of invitation and information booklet which explained the process involved, and an easy-to-read appointment card (see appendix 13 for *Health Fair* invitation pack). Each assessment was also broken down into 4 to 5 steps with simple plain language instructions per step and a pictorial representation (see appendix 8: 35). These steps reflected prior consultation with people with ID and were consistent with the participatory approach employed. See *figure 7.1* for the multiphase approach adopted for the *Health Fair* process development,

which is in keeping with and influenced by the underpinning Values Framework (McCarron *et al.*, 2011) of the IDS-TILDA study (see Chapter 4: 74).

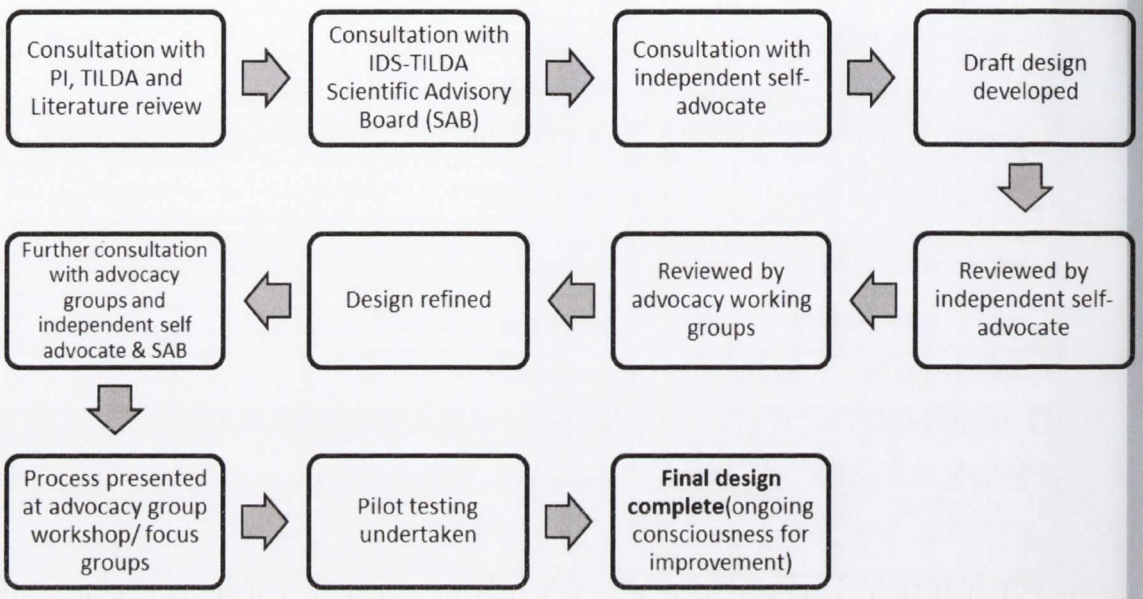


Figure 7.1: Multiphase Health Assessment & Accessible Material Development Process

Initially, the assessments to be included in the *Health Fair* were identified and as far as possible replicated those non-invasive assessments included in the TILDA study. The literature was reviewed and the IDS-TILDA PIs were consulted to ascertain scientific rationale and accurate information on the assessments. Consultations occurred between the researcher and the health assessment team of TILDA. The researcher also presented the proposal to the scientific advisory board of IDS-TILDA for further advice, further the researcher consulted with an independent self-advocate with regards the inclusion of the particular assessments. Following this, draft health assessment accessible information and support materials were developed and again reviewed by the independent self-advocate.

The researcher contacted several advocacy groups and presented the material to them for critique and feedback which contributed to refining the design. Any issues that arose were discussed with the varying groups and solutions identified and employed. **Table 7.3** presents the issues that arose and the amelioration strategies employed during the development and consultation process. The health assessment process was then

presented to a broad team of advocates for final revision and feedback before the pilot study commenced. Throughout the consultation and review process all suggestions and revisions were considered and incorporated. The pilot study further contributed to refining the process.

Table 7.3: Issues and solutions in designing an accessible model of delivery for IDS-TILDA *Health Fair*

Issue	Identified by	Solution
Comprehension of participants	Scientific Advisory, Advocacy groups, PIs, Independent experts, Independent advocate, Researcher	Development of a communication strategy to incorporate • Easy read information booklet prior to day of assessment • Liaison with service link personnel • Identify need for key worker presence to support participant and clinical researcher • Introduction of alternate modes of communication – hand over hand; Lámh; visual demonstration • Enlarge explanatory pictures • Encapsulate explanation on one page with no more than 4-5 steps
Obtaining consent	Scientific Advisory, PIs, Independent experts, researcher & Service link personnel	• Development of easy read consent form • Ongoing process consent employed throughout assessments • Collaboration with key support personnel before and during assessment especially for those participants who were non verbal • Careful observation of body language of participant
Inclusion of non-ambulatory participants	Scientific Advisory, PIs, Independent experts, researcher & Service link personnel	• Inclusion of alternate methods of measuring height and BMI – use of the MUST tool • Maintain accurate records regards ambulation
Identification of results outside normal parameters	Scientific Advisory, PIs, Independent experts, researcher & Service link personnel	• Develop easy read results card • Identify results and explain to participant • Highlight possible follow up with their GP
Privacy and participation issue	Scientific Advisory Service link personnel & Advocacy groups	• Emphasise voluntary nature of participation • When obtaining consent highlight right to withdraw at any time • Employ emancipatory principles • Acknowledge participant as leader at all stages of assessment
Properly prepared for assessment	Advocacy groups	• Ensure full instructions are relayed to participants prior to assessment • Provide contact details for any queries • Allow and ensure sufficient contingency time is built into total process • Let the participants know specifics required prior to arrival at assessment e.g. no tights, light layers of clothing

7.4 Pilot study

The researcher undertook a pilot study with a purposive sample of 25 individuals with various levels of ID, from a variety of living circumstances aged 40 years and over. All participants were independent of the main study, see **table 7.4** for demographic profile of pilot study.

Table 7.4 Demographic profile of pilot study participants

Demographic Variable (N=25)*		Frequency	%
Age	40-49	3	12.5
	50-59	10	41.7
	65+	11	45.8
Gender	Female	18	75.0
	Male	6	15.0
Level of ID	Mild	13	54.2
	Moderate	9	37.5
	Severe	2	8.3
Residence	Independent/semi	8	33.3
	Community group home	14	58.3
	Residential setting	2	8.3

**One participant would not engage on the day.*

The pilot study assisted the researcher in identify potential problems and therefore rectify them before commencing the main data collection. It also provided an opportunity for participants to give feedback on their experience of engaging in the health assessment process. The researcher included a feedback form with a section for open narrative on opinion to ascertain positive and negative aspects of the study which would further contribute to improving the overall experience for the main study participants (see appendix 14). See **table 7.5** for the opinion ratings of each of the assessments. The pilot study also demonstrated the willingness of people with an ID to assist and give their opinion to improve the experience for others.

Table 7.5: Overall feedback on opinion of pilot participants (N=25)

Assessment Type	Rating			
	Liked it a lot *n* (%)	It was Ok *n* (%)	Did not like it *n* (%)	Unable to answer *n* (%)
Grip Strength	17 (70.8)	5 (20.8)	0	2 (8.3)
Waist to Hip ratio	15 (62.5)	7 (29.2)	0	2 (8.3)
Blood Pressure	11 (45.8)	10 (41.7)	1 (4.2)	2 (8.3)
Height	15 (62.5)	7 (29.2)	0	2 (8.3)
Weight	14 (58.3)	7 (29.2)	0	2 (8.3)
Timed up and go	14 (58.3)	8 (33.3)	0	2 (8.3)
Quantitative Heel Ultrasound	17 (70.8)	5 (20.8)	0	2 (8.3)

The pilot study also served to provide a method for identifying the length of time it would take for setting up, quality checking and calibrating the assessment equipment and for timing the process, in a variety of settings and identifying potential challenges that may occur when out in the field. The completion time ranged between 10 - 50 mins. The lower time occurring for a person who did not engage in all elements of the health measures. The pilot study gave the researcher insight into improving the flow of the process, the efficiency of where to position equipment, the importance of space, the efficacy of the easy read explanatory materials and the challenges that lay ahead in the main study if the assessments were to take place in a person's home. Identified factors that contributed to a longer completion time included severe/profound level of ID, immobility status, difficulty with communication and the presence of other factors such as challenging behaviour. And finally, the pilot study demonstrated that doing the assessments on all participants is possible once certain key elements are in place, such as a familiar support person for reassurance of the participant and guidance for the researcher.

7.5 Quality Assurance

The researcher also endeavoured to maintain quality assurance throughout the total process from inception to dissemination. To that end the researcher developed an audit process with identified measures, procedures to employ and outcomes or deliverables to meet. This procedure ensured the researcher remained cognisant at all stages of the research process of quality in her actions and procedures. **Table 7.6** enumerates and presents the process employed.

Table 7.6: Evaluating and Maintaining Quality Assurance Process

Audit Process	Measure	Procedure	Deliverables
Planning	Development of *Health Fair*	Review by Scientific Advisory Committee, Advocacy consultation groups and independent experts	Health Assessment Protocol, accessible focus group consultation presentation and accessible demonstration materials developed
	Development of accessible material	Review by Scientific Advisory Committee, Advocacy consultation groups and independent experts, pilot test	Easy read consent, easy read assessment explanatory material, easy read appointment and information booklet, easy read results card
	Expertise of researcher	Assessed by expert peers, engagement in further & specific training, engagement of people with ID to review and review by supervisors	Demonstrate competency in health assessments, completion of the 'Osteoporosis Essentials Course', overall roll out of *Health Fair* and engagement of people with ID
Performance	Roll out of *Health Fair*	Pilot study	Assessment of feedback from pilot study participants
	Promotion of unique experience	All materials reviewed by experts, weekly/fortnightly updates reports of progress, meticulous planning and project management	Ensuring sufficient information prior to appointment, sufficient timing of appointments, key workers accompanied participant, individual appointments with sufficient time for each participant
	Data collection quality	Refresh training in clinical procedures of health assessments Development of monitoring and recording system Liaison with equipment service personnel Maintenance of daily records Include a process of continuous evaluation of procedure	Attend the TILDA health assessment clinical nurse advisory for induction into health assessment procedure Attend Clinical Nurse Specialist at a large national specialist bone unit for assessment of technique. Accurate and on the spot data recording in specifically designed data collection form, maintenance of equipment, cleaning, testing and calibration. Record accurate records of successes and difficulties, enumerate solutions to difficulties
	Data entry quality	Enter data as soon as possible post assessments Quality assurance and cleaning process applied to complete dataset, sample checked for errors, independent researcher checks	Clean dataset

Table 7.6: Evaluating and Maintaining Quality Assurance Process (continued)

Audit Process	Measure	Procedure	Deliverables
Reporting	Data analysis quality	Completion of quantitative analysis course, independent data analyst cross checking results, consultation with statistician regards analytic methods applied for quality check, consultation with research team	Accurate clear results
	Dissemination	Through the identification of high quality peer review conferences and publication. Strategic sharing of information with service providers, staff and service users Consultation with supervisors	Results disseminated in a timely and efficient manner appropriate for the audience intended

(From inception to dissemination & ongoing)

7.6 Statistical Analysis

For the purposes of this analysis the statistical analysis was conducted using the Statistical Package for Social Sciences (SPSS) version 22 (IBM 2013). All data entered was cleaned through a process of preliminary descriptive analyses identifying frequencies, distribution and identification of missing values. With consideration to missing data valid percentages only are presented throughout the paper. Independent t-test were carried out on continuous variables to assess the differences between the age group categories of those who attended and the non-attenders of the *Health Fair*, to explore the relationship between categorical variables Pearson's chi-squared test of association was used.

All verbatim text from the field notes, observations and direct quotes were exported into an excel spreadsheet for thematic analysis. The excel sheet allowed colour coding, grouping and filtering of all the data. Braun & Clarke's (2006) framework was applied. This entails a six step approach:

1. Familiarising yourself with the data
2. Generating initial codes
3. Searching for themes
4. Reviewing themes
5. Defining and naming themes
6. Producing the report

This provided the opportunity to extract examples and select extracts to support and relate back to the point under scrutiny. Thematic analysis of the text allowed key commentary and opinion on the participant's experience of the *Health Fair* to be extracted.

7.7 Results

Of the 753 participants who were interviewed nationally for Wave 1 of the IDS-TILDA study [2011], 5.9% withdrew or were deceased prior to Wave 2 commencing. The remaining 94% completed the Wave 2 computer assisted personal interview (CAPI) and were invited to participate in the objective health assessments. Of those, 86.3% (n=611) agreed to participate, however 7 participants withdrew on the day of the assessments, leaving a total uptake of 85.3% (n=604). Of those who participated 50.8% (n=307) engaged in all elements of the *Health Fair* and 49.2% (n=297) engaged in at least one element of the *Health Fair*. Of those who did attend 47.1% were supported to complete their assessment with the assistance of their support staff/carer. In total, 14.7% (n=104) did not participate in the *Health Fair*, see *figure 7.2* for overall details and details given for nonattendance.

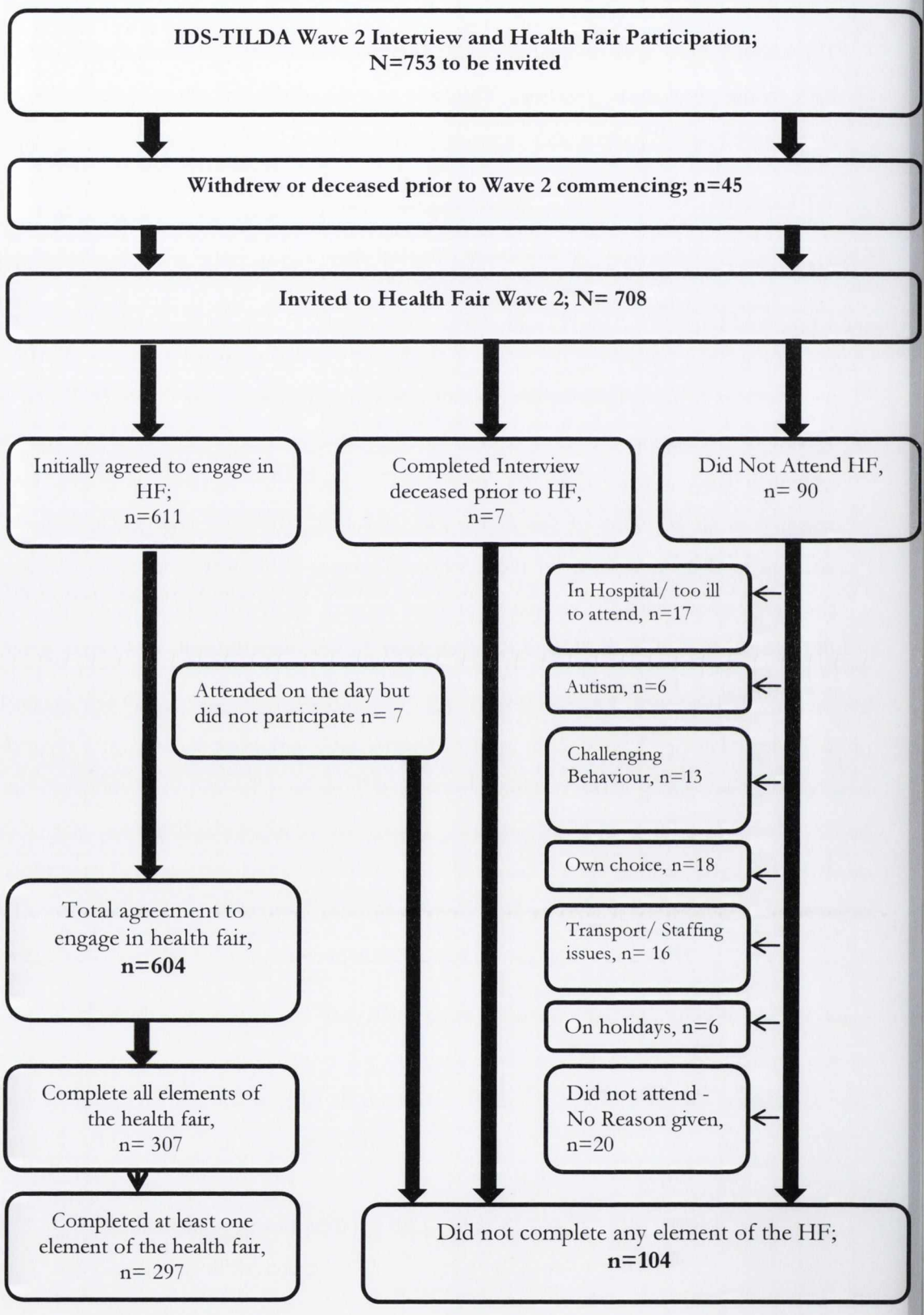


Figure 7.2: Health Fair Attendance Flowchart

Table 7.7 details the sociodemographic, physical health and behavioural characteristics of the participants who did and did not attend the health assessments. With regard to three demographics referred to here, those who did and did not attend varied significantly with regard to gender (*p*-value = 0.009) but not regards to age and level of ID.

Table 7.7: Sociodemographic, behavioural and physical health characteristics of participants in the IDS-TILDA

Characteristics*		Did not attend assessment (n= 104)	Attended assessment (n=604)	Totals	N	*p-value*
Sociodemographic						
Age	Mean ± standard deviation	57.50 ± 10.06	56.44 ± 9.18	56.59 ±9.31	701	0.283[4]
Gender	Male, n (%)	58 (55.8)	254 (42.1)	314 (44.1)	708	**0.009**[2]
Level of ID, n (%)	Mild	29 (31.9)	129 (22.9)	158 (24.2)	654	0.177[5]
	Moderate	37 (40.7)	267 (47.4)	304 (46.5)		
	Severe/Profound	167 (29.7)	25 (27.5)	192 (29.4)		
Physical and Behavioural Health						
Self-rated health, n (%) (excellent, v. good, good)		72 (72.0)	508 (86.4)	580 (84.3)	688	
Barthel ADL Index⌘, n (%)					696	
	Independent	18 (17.6)	93 (15.7)	111 (15.9)		
	Slight dependence	24 (23.5)	100 (16.8)	124 (17.8)		
	Moderate dependence	14 (13.7)	109 (18.4)	123 (17.6)		
	Severe dependence	46 (45.1)	292 (49.2)	338 (48.6)		
Presence of Mental Health condition, n (%)		42 (42.4)	304 (52.8)	346 (51.3)	675	
Has Challenging Behaviour, n (%)		69 (73.4)	418 (74.0)	487 (73.9)	659	
Physical Activity level (low), n (%)		75 (74.3)	431 (73.1)	506 (73.2)	691	
Communication (Difficulty), n (%)		62 (60.8)	363 (61.2)	425 (61.2)	695	

*Obs missing (not all participants answered all items)

⌘ (Wade and Collin 1988) Barthel ADL Index (modified: see appendix 15); ADL=activity of daily living

p-value set at 0.05, statistically significant results in bold

[4] t-test for equality of means

[5] Chi-Square Test for Association

Table 7.8 summarises the range of scores including mean and median for the objective health measures completed in the *Health Fair.*

Table 7.8: Objective health measures

Health measure*	Mean	±SD	Range (max-min)	Median	Interquartile range	Total
Height (cms)	159.5	10.8	190-103	159	14 (153-167)	594
Weight (Kgs)	72.7	15.1	131.1-37.9	71.5	20 (61.85-81.85)	492
Waist circumference (cms)	100.0	13.2	136.0-63.5	99.5	17.6 (0.89-0.99)	514
Waist to hip ratio	0.94	0.08	1.34-0.69	0.94	0.1 (0.89-0.99)	513
Timed up and go (total seconds)	17.1	13.8	222.8-5.9	13.7	8.2 ((10.74-18.96)	490
Hand grip strength (Kgs)	17.4	8.8	50-1	16.5	11.25 (11.5-22.75)	473
Blood pressure (systolic)	118.8	18.1	192.0-77.5	117.5	23.5 (106-130)	551
Blood pressure (diastolic)	75.8	12.2	117-46	75	15.5 (68-83)	551

*Not all participants who attended the health assessments (n=604) completed all the measures

Table 7.9 presents the results amenable to standard classification and enabled the categorisation shown. The QUS calculated the speed of sound (SOS) and broadband attenuation (BUA) to the stiffness index (SI). This was then further presented as t-scores with referenced to the mean average healthy European young adult indicating the risk categories of fracture. The QUS t-scores enabled the classification as described above. Grip strength was stratified by age and gender according to the manufactures instructions.

Table 7.9: Objective Health Measure Outcomes

	Categories	*n* (%)	Total (n)*
Bone Quality QUS	Normal	148 (25.7)	575
	Osteopenia	191 (33.2)	
	Osteoporosis	236 (41.0)	
BMI	Underweight	20 (3.4)	592
	Normal	177 (29.9)	
	Overweight/Obese	395 (66.7)	
Waist to hip ratio	Excellent/good	45 (8.8)	513
	Average	111 (21.6)	
	At risk	357(69.6)	
Grip strength (stratified by age and gender)	Below	418 (93.3)	448
	Normal	16 (3.6)	
	Above	14 (3.1)	

**Not all participants completed all measures*

As has been noted, not all potential participants completed the health assessments. As may be seen in **table 7.10** gender and self-rated health are significantly associated with attendance at the health assessments. Females are significantly more likely to attend the health assessment than males (88.4%, n = 396 vs 81.4%, n=312; OR=1.737, 95% CI

=1.142 – 2.643). Those with self-rated excellent/v. good/good health are significantly more likely to attend than those with fair/poor self-rated health (OR=2.469, 95% CI=1.504 – 4.056), 87.6% (n=580) vs 74.1% (n=108).

Table 7.10: Associations between participant characteristics with attendance or non-attendance at health assessment

Characteristics		Crude Odds Ratio (95% Confidence Interval)	p-value
Gender (Reference male)		1.737 (1.142 – 2.643)	**0.010**
Age (Reference 44 – 49)			0.420
	50 – 64	0.963 (0.582 – 1.595)	0.884
	65+	0.708 (0.395 – 1.266)	0.244
Level of ID (Reference mild level of ID)			0.177
	Moderate	1.622 (0.955 – 2.755)	0.073
	Severe/Profound	1.502 (0.839 – 2.688)	0.171
Poor self-rated health (Fair/Poor)		2.469 (1.504 – 4.056)	**<0.0001**
Barthel ADL Index* (Reference independent)			0.295
	Slight dependence	0.806 (0.411 – 1.581)	0.531
	Moderate dependence	1.507 (0.711 – 3.194)	0.285
	Severe dependence	1.229 (0.679 – 2.223)	0.496
Physical activity (Reference low)	Moderate /High	1.064 (0.657 – 1.723)	0.800
Mental Health condition (Reference no MH)		1.517 (0.986 – 2.334)	0.058
Challenging Behaviour (Reference no CB)		0.993 (0.877 – 1.123	0.906
Communication difficulty (Reference no difficulty)		1.018 (0.662 – 1.566)	0.934

**Modified Barthel Index (see appendix 15)*
p-value set at 0.05, statistically significant results in bold

7.7.1 Identifying contributions and challenges to success on the day of the *Health Fair*

Braun and Clarkes (2006) thematic analysis framework was employed to analyse the observations, field notes and comments from participants. Through this technique the commentary was grouped and classified into themes to enable identification of positive and negative feedback from the day of the *Health Fair.* In total there were 512 observations, comments and/or feedback from participants. This represents 84.8% of the total *Health Fair* participants. The main themes to emerge were categorised into two global themes, the promoters which contributed to the success on the day and the inhibitors which posed challenges on the day (see *figure 7.3*). The promoters to emerge included staff support, enjoying the interaction and experience on the day and receiving acknowledgement for their contribution and achievement (see appendix 16 for certificate

of achievement). Potential inhibitors included being anxious or nervous about what was going to occur, behaviours that challenge, communication challenges and physical health or mobility issues. These comments were documented throughout the *Health Fair* on a daily basis and enabled the researcher examine not only what she was doing that promoted participation but also provided experiential learning of how the potential inhibitors were ameliorated, how the process was amenable to the participants and how, by ensuring the key fundamentals of the underpinning philosophy were maintained, the outcome was more favourable for participants. The thematic analysis enabled identification of feedback from the participants in support of the findings as discussed below.

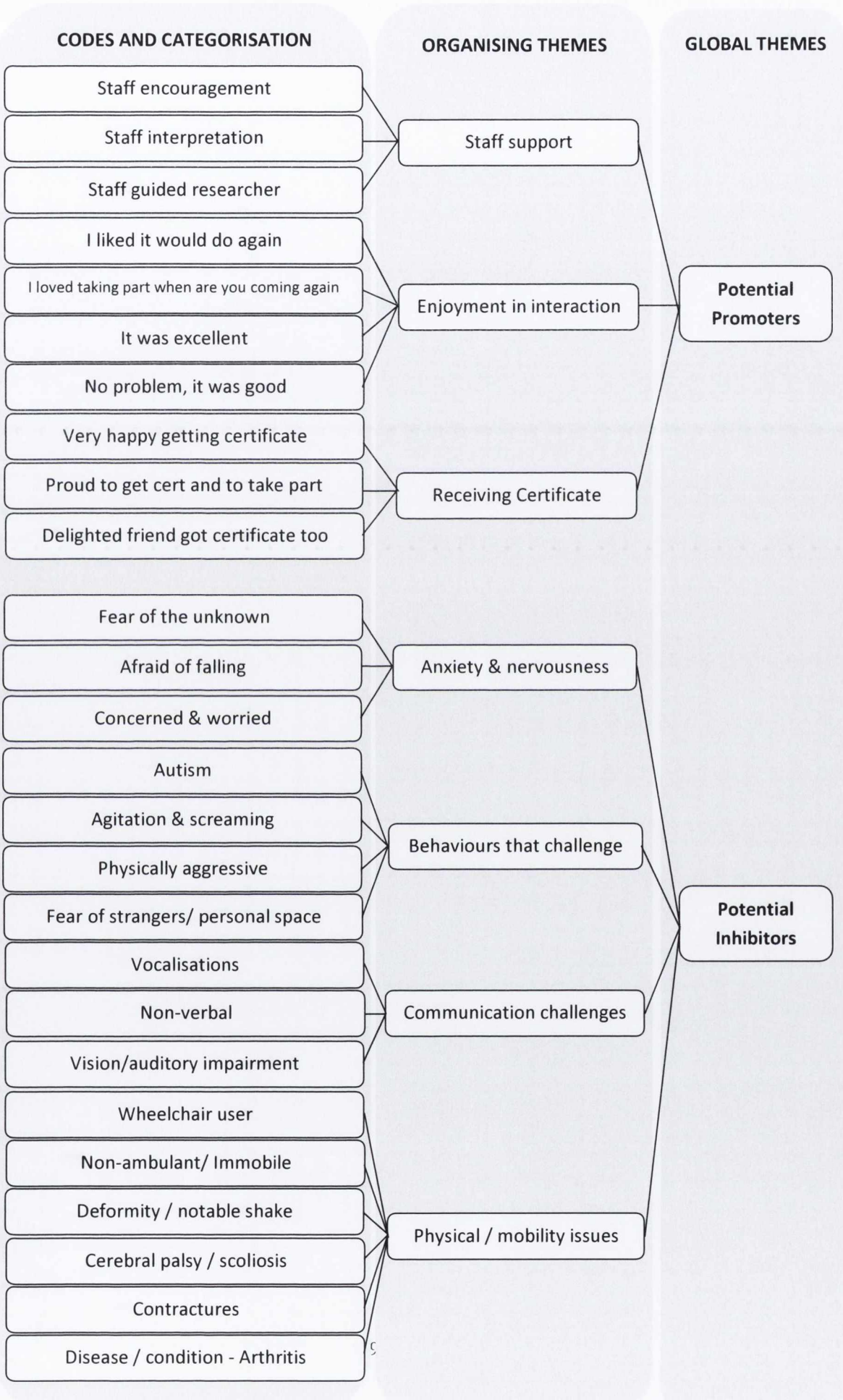


Figure 7.3: Potential inhibitors and contributors to participant engagement with the Health Fair

7.8 Discussion

Reflecting the implementation of a successful process over 85% of the invited participants engaged in the Health Fair



Despite the current increase in lifespan, people with ID continue to die earlier (McCarron *et al.,* 2015), present with more complex and adverse health conditions, experience more health challenges (McCarron *et al.,* 2013a, McCarron *et al.,* 2013b, Krahn *et al.,* 2006) and inequalities engaging in health care and health promotion, with needs often going unmet (Lennox and Kerr 1997, Robertson *et al.,* 2011). IDS-TILDA is concerned with improving the health and well-being of all people with ID as they age. Ageing among people with ID is notable in Ireland (Kelly and O'Donohoe 2014) however the mortality rates continue to be higher than those of the United States or Australia for example (Janicki *et al.,* 1999, Bittles *et al.,* 2002). The absence of objective measurements of health for people with ID from intellectual disability population studies adds to the lack of health information about this cohort and contributes to marginalising this group of people. To that end, including objective health measures as part of the IDS-TILDA study to assist in the development of a deeper understanding of the ageing process of people with ID was essential. This challenging task involved the collaboration of a multidisciplinary team; the participants, carer's, service providers, advocacy groups and experts in the field of ID. The success of this current study whereby over 85% (n=604) of the IDS-TILDA participants engaged in health assessment demonstrates that it is possible to obtain quality objective measurement with people with ID.

7.8.1 The health profile

Of the males, 81.4% (n = 312) attended the *Health Fair* and of the females 88.4% (n= 396) attended. Although the difference is typical of society whereby females are more likely to be concerned with preventative health, conversely men, as they get older, are more likely to present with a pattern of reactive healthcare. Of those with mild ID 81.6% (n = 158) attended, of those with moderate ID 87.8% (n = 304) attended and 87% (n= 192) of those who attended reported a severe/profound. Of those who engaged in the QUS assessment the majority presented with bone compromise and although the QUS cannot be used on its own to diagnose osteoporosis it is predictive of hip fracture in older women, here over

70% had indications of either osteopenia or osteoporosis. The anthropometric measures of waist to hip ratio (WHR) and BMI estimate the fat distribution of the body and are commonly utilised as indicators of risk for cardiovascular events. In this study it can be observed that the majority of participants are within the overweight and obese BMI category (66.7%) and further, the WHR of 69.6% of participants was within the 'at risk category'. Overall this indicates the potential for poor cardiovascular health among participants and a reflection of a sedentary lifestyle.

7.8.2 Attending or not, what are the determinants?

There was very little difference in the physical health profile of those who attended and those who did not attend the *Health Fair*. Significantly more females attended than males however it is important to note that there are more females in the study overall however typically in society women are more likely to attend to their health needs more than men. More people who attended reported their health as excellent very good or good 86.9% versus 72% of those who did not attend. This is not surprising considering that one of the concerns expressed by those that did attend as potential inhibitors was physical or health issues and this was also identified on examining the reported reasons for non-attendance. One participant was restricted in the number of assessments they could participate in, assessor noted '*...is in a lot of pain when standing, found it too difficult, c/o pain +++; currently awaiting total hip replacement*'. Another example of physical health restricting engagement is demonstrated from the following extract when one participant expressed how painful movement was for him due to his arthritis; assessor noted '*any movement causes pain and distress, arthritis very painful*'. To note, a greater proportion of participants who did attend were more likely to have some level of dependence as identified through the Barthel ADL Index, this possibly is reflective of the fact that those who did require assistance were supported to attend by their carer's or support staff. More people with mental health conditions attended with very little difference between those who attended and did not attend when examining factors like challenging behaviour, physical activity or communication difficulty, again perhaps reflective of the support they received to attend however the differences were not extensive between the two groups, see for example Barthel Index score - severe dependence; 45.1% non-attenders versus 49.2% attenders. The picture that emerges on examining the reported reasons for non-attendance indicates that of those who did not attend more often did so of their own choice, had difficulties with behaviour which prevented them from attending or did not give any reason at all.

However there were a small number of issues that could possibly be addressed to support the person to attend, such as lack of transport, staffing issues or being away on holidays on the day of the *Health Fair.*

7.8.3 Contributors to success

Nevertheless the percentage of engagement in the *Health Fair* indicates the overall success and achievement of the participants, their carer's and the *Health Fair* process itself. An important element in this success was underpinning the whole process with the key fundamentals of an emancipatory philosophy. This provided the framework through which an infrastructure could be built to make possible the application of novel approaches to each of the assessments and so improve participant involvement, create processes with reasonable adjustments and ultimately promote success. The key fundamentals complement and work together to promote overall reasonable adjustment of each of the assessments, therefore ensuring the whole process, to a great extent, meets the needs of each participant.

Empowerment ensured the sharing of the decision-making through the collaborative process working with advocacy groups through to ensuring the participant on the day of the assessments retained full independence in their decisions about taking part. The accessible consent was thoroughly explained with the support of pictorial representation. One staff attending commented *"I'm 22 years working in this service area and that's the first time I've seen a well explained user friendly procedure with anyone with ID".* The underpinning framework also ensured information was in a form understandable to all stakeholders and promoted participant ability to enable each attain their potential to take part through the use of easy-read information and explanatory material. This was demonstrated through this participants comment, *"Ah, I'd stay here all day that was easy and lovely".* Empowerment was supported through the involvement of the participant's carer and service provider and the application of multiple communication methods to ensure full understanding, such as the use of Lámh, hand over hand, pictures, demonstrations and carer's demonstrating, all to build confidence with the participant in the process.

The carer's support on the day was imperative, their knowledge of the participant, their support and encouragement and their advice to the researcher contributed greatly to a successful *Health Fair.* Complimenting empowerment in the emancipatory philosophy is the second key element namely reciprocity, ensuring respect and dignity to all participants.

The researcher ensured choice was respected at all times, for example, although the lay out of the *Health Fair* was specific as to reduce burden, it also needed to be flexible enough to ensure it met the choice and interest of the participant. Therefore if a participant chose to start with having their blood pressure measured, this was facilitated. Building capacity was essential to participant success therefore including surrogate measures and developing alternate techniques and approaches to assessments, such as the technique for mounting the standiometer to measure height or, the distraction technique employed when taking blood pressure measurement to reduce anxiety, were all invaluable. People expressed their satisfaction of the process and enjoyment of taking part indicating that they would not mind doing it all again, another reflection of a successful process. These sentiments are best expressed by participants who said *"It was good it was excellent and I wouldn't mind doing it again"* and another who said *"Ah good, I liked it, when are you coming again"*.

The final elements to the emancipatory philosophy are social engagement and gain. These were operationalised through offering the assessments at a site convenient to participants where possible, ensuring the participant and their carer's were furnished with information prior to the *Health Fair*. This material informed them of the study and the potential contribution to the health and well-being of people with ID. Providing the results and explanations in an easy-read format and highlighting any anomalies for further action. This was appreciated by staff and participants as some opportunities such as having a bone assessment may not have occurred previously. The following quote by one of the participants sums up the value succinctly *"doing things like this, you get to know what your health is like"* with another adding *"tis lovely, it's great to get all them things done I'm delighted"*. Further it was important to acknowledge the participant's involvement and achievement. Participants frequently conveyed their delight in taking part, for example one participant said *"I loved it, I just loved it. I loved everything"*. The participants also received a certificate of achievement on completion of any or all of the *Health Fair*. The participants expressed pleasure and enjoyment in receiving this, demonstrated by this quote from a participant *"Oh that'll go up on my wall so it will - I'll get a wee frame for that"* and frequently told their friends or proudly displayed their certificate in their homes. Sometimes it was the delight in receiving the certificate that instilled a pride in their participation. For one participant support staff noted that anything new or different created anxiety and the participant became very scared, however working through the *Health Fair*, employing all techniques and promoting relaxation the participant

completed all elements and was thrilled when he received his certificate. The participant was excited and could not wait to go to tell all the staff about what he had achieved.

7.8.4 Overcoming challenges

The emancipatory philosophy also contributed to overcoming many of the challenges encountered throughout the *Health Fair* process. Ensuring easy-read documentation and alternate augmented communication helped overcome communication barriers and difficulties with comprehension along with support from key staff and carer's. Using techniques such as demonstration and hand over hand, elucidated procedures for people building on their understanding and making the whole process easier. Employing welcoming strategies, ice breakers and observational techniques to allay fears and anxiety reduced people's reluctance to engage and promoted confidence. This was portrayed when one participant attending appeared quite restless and the accompanying staff noted that it would be difficult to get him to cooperate, however with careful explanation the participant smiled when getting the QUS done and staff noted that he had enjoyed that and they were surprised as to how many assessment the participant completed. On another occasion staff expressed their surprise that another participant had stayed calm as the participant's usual demeanour was distress and agitation.

In an attempt to anticipate and deal with any potential environmental barriers to participant engagement in the *Health Fair*, the choice of room and space requirements were negotiated before the *Health Fair* appointments, confirming accessibility to the *Health Fair* room and ensuring adequate signage further addressed such potential challenges. Employing innovative techniques to make participation easier and more comfortable along with using alternate measures culminated in making the *Health Fair* as accessible as possible and suitable for all participants.

7.9 Limitations

Some participants did not engage in all of the assessments, a proportion of participants, especially those with behaviours that challenge and autism had particular difficulty despite employing strategies to engage the participants. It is noted that perhaps with time these particular participants would build familiarity with the processes and assessments and could successfully engage in future. For people with severe to profound ID, grip strength and the timed-up and go test, for which there were no alternatives, posed particular

challenges. However, there was a good overall response rate and every effort was made to encourage and facilitate participation.

7.10 Conclusion

In summary this article details the assessments, approach, philosophy and structure adopted in undertaking objective measurement with people with ID. Frequently it is said that engaging people with ID in research and especially research which involves objective measurements are too difficult, too stressful for the person with ID or just not possible. As can be seen by the success of the process implemented in this study these excuses are not acceptable and measures ought to be taken to include people with ID. Excluding people on these grounds is colluding in the disenfranchisement of people with ID. As demonstrated in this study with innovation, adoption of adaptable and flexible guidelines embedded in an emancipatory philosophy, inclusion of measurement is possible for people with ID and ensures '*nothing about me, without me*' (Delbanco *et al.*, 2001) resonates as the pinnacle of shared decision-making in carrying out the assessments in a collaborative manner preventing the sole focus being merely a measuring exercise. Critically this study provides an evidence based procedures that could easily be transferred to a clinical setting to assist not only the research community but more importantly the doctors, nursing and multidisciplinary team to ensure equity of access to health care for people with ID.

Further specific detail is available in the '*The Health Fair Protocol* (appendix 8). It describes the complexities involved in the health assessment process. It is hoped that this work will provide the design from which other researchers and clinicians can draw and develop processes of engagement, enabling people with ID be true partners in research and an improved healthcare delivery system. With the right processes and enablers in place people with ID of all levels and abilities can engage in objective measurements to provide and contribute to building a robust picture of their health as they grow older. In doing so, contribute to developing a greater understanding of their health pathways to improve their health care delivery and services now and to the future.

This chapter has presented the overall process, procedure and participation of the *Health Fair.* The following chapters focus on the GE Lunar Achilles bone health measurement and results. This chapter has identified how, by employing a collaborative

and augmented process health screening is possible and particularly bone health. An exploration of the results of the bone health screening is presented in the next chapter.

CHAPTER 8

EXPLORING THE OBJECTIVELY MEASURED BONE STATUS OF ADULTS WITH INTELLECTUAL DISABILITY

"Take care of your body it's the only place you have to live"

- Jim Rohn

Chapter summary

The following chapter examines the results of the objectively measured bone status of the participants in the IDS-TILDA study Wave 2 [2014]. Overall the prevalence of osteoporosis among people with ID in Ireland has been reported at much lower levels than the general population, however objective evidence from the general population study (TILDA) identifies noticeable discrepancies between the self-reported doctor's diagnoses and objectively measured levels of osteoporosis. In the second wave of the IDS-TILDA study objective measurement of bone health was obtained using quantitative ultrasound of the calcaneus (QUS). People also reported if they had a doctor's diagnosis of osteoporosis. Objective evidence of poor bone health is identified at over 70% in this study, with a higher prevalence among women than men, and a definitive age gradient noticeable. Those with more severe/profound level of ID are more likely to present with osteoporosis. However, what is concerning is that men have higher levels of fracture rates, are less likely to be screened and are 12 times more likely to present with objective evidence of osteoporosis than their general population peers. The picture is especially bleak for men with Down syndrome where 100% of them did not have a doctor's diagnosis when considering the findings of the objective evidence in this study. The high levels of objective evidence of poor bone health identified among this cohort increases the risk of fractures as people age. Prevention strategies for both osteoporosis and fracture in this population should be a priority.

8.0 Introduction

Osteoporosis is a progressive bone disease characterised by low bone mineral density (BMD) and microarchitectural deterioration, leaving the bone susceptible to fragility fracture (Kanis *et al.,* 1994). It is becoming one of the most common diseases associated with ageing as the older generation increases exponentially and, is of major public health concern because of the serious consequences in terms of morbidity and mortality (Beard 2011). Fracture of the hip, wrist or vertebrae are recognised as typical osteoporotic fractures (Kanis *et al.,* 2009a) and commonly occur as a result of very low trauma events. However, these events can impact detrimentally on a person's life. The European figures of one third of women and one fifth of men over 50 having a diagnosis of osteoporosis are not evident from HIPE[6] data in Ireland (Gannon *et al.,* 2007). However, it is estimated that over 300,000 people in Ireland have osteoporosis and due to the silent nature of the condition many are unaware (Health Service Executive 2008). In Ireland there is no national policy on osteoporosis (HSE 2008). A national strategy group reported in 2008 on the national costs and figures for falls and fractures and noted that annually, approximately 7000 older people present with fracture, most probably as a result of osteoporosis. These events have an annually costing in excess of approximately €59million, 80% of these events were in the over 75 year age group most of whom suffered hip fracture, one of the major osteoporotic fracture (Gannon *et al.,* 2007).

For people with intellectual disability (ID) who frequently presents with many health challenges, the risk of undiagnosed osteoporosis and risk of fracture is possibly very high (Angelopoulou *et al.,* 2000, Srikanth *et al.,* 2011, Jasien *et al.,* 2012). Health challenges such as co-morbidity, communication difficulties and very often unmet health needs (Krahn *et al.,* 2006, Lennox *et al.,* 1997, Cooper *et al.,* 2004), contribute to this higher risk. Studies among people with ID investigating osteoporosis have relatively been restricted to small population sizes (Arvio *et al.,* 2009, Center *et al.,* 1994, Center *et al.,* 1998). However, Zylstra *et al.,* (2008) investigated the prevalence of osteoporosis in a larger group of adults with ID living in the community (N=298) and identified the rate of osteoporosis of the femur at 17.1% and osteopenia at 51%. Overall the study identified a mean t-score of -1.71

[6] The Hospital In-Patient Enquiry (HIPE) scheme, established in 1971, is a health information system designed to collect clinical and administrative data on discharges from, and deaths in, acute hospitals in Ireland. It is the principal source of national data on discharges from acute hospitals (HIPE 2013).

for all ages, both males and females. Of note in this study was the higher prevalence identified for males, 19.2% versus 14.5% respectively. Other studies vary from only 1% of an entire sample having a pre-existing diagnosis of osteoporosis (Tyler *et al.*, 2000) to 8% in the Irish ID population study (McCarron *et al.*, 2011). Subsequently in the Tyler study, following measurement, over 21% of participants were identified within the osteoporotic range and a further 35% were found to be osteopenic (Tyler *et al.*, 2000).

People with ID have very specific health needs and frequently these are not taken into consideration when policy or strategy is being formulated. This increases the potential for conditions such as osteoporosis going unrecognised among this cohort. Contributing to the problem, along with the previously mentioned challenges are difficulties many people with ID have when it comes to diagnosis and measurement (Lennox *et al.*, 1997). Diagnosis can be fraught with difficulty as people with ID may not be able to express their symptoms, may fail to report symptoms or may find the assessment and diagnostic process, dual energy X-ray densitometry (DXA), frightening and intimidating (Kirschner *et al.*, 2007). On the other hand they may not tolerate this method at all due to anatomic deformities or behavioural presentation. Lohiya *et al.*, (2004) reports that almost 40% of the participants in their study were unable to have BMD measured because of physical and behavioural issues. Despite the known challenges of assessment and the sometimes unsuitability of DXA there is no robust assessment protocol for people with ID, alternate screening methods are rarely utilised and many people with ID continue to go without proper bone health assessment (Srikanth *et al.*, 2011, Michael 2008). Over the last number of decades new technology has been developed for the evaluation of skeletal health namely quantitative ultrasound (QUS). These devices are low-cost, radiation free with the measurement complete in 10 seconds. As an alternate method of bone health screening the QUS can give an indication of bone quality and contribute to identifying the risk of fracture, and, combined with clinical risk factors can contribute to implementation of prevention or treatment (Njeh *et al.*, 1997, Hans *et al.*, 2003).

8.1 Quantitative Ultrasound

Establishing and identifying people at risk of osteoporotic fracture reflects a comprehensive clinical approach to osteoporosis management. Dual-energy X-ray absorptiometry (DXA) particularly of the femoral neck is the 'gold standard' and is commonly used in the identification and classification of osteoporosis and osteopenia (low

bone mass) incorporated with widely used clinical risk factor assessment tools such as FRAX, it is also utilised for monitoring any changes that may occur in bone health status overtime (Kanis *et al.,* 2009b, Kanis 2002, Kanis *et al.,* 2008a, Kanis *et al.,* 2008b). The International Society of Clinical Densitometry (ISCD) has established an official position on QUS in the management of osteoporosis (Krieg *et al.,* 2008). They note that the use of QUS is justified in the absence of DXA however they acknowledge there are technological diversity between DXA and QUS, and between the varieties of QUS devices that are available. Subsequently, the results for each cannot be extrapolated from one device to another for equal comparison. The ISCD highlight that the only validated skeletal site recommended for ultrasound scan is the os calcis (heel).

It is also noted that QUS has a practical role to contribute in the identification of those at risk of osteoporotic fracture in conjunction with clinical risk factors. The ISCD emphasise that validated QUS devices predict fragility fracture independently of DXA BMD (Krieg *et al.,* 2008). The instrument utilised by the ISCD as their reference device is the GE Lunar Achilles however it is important to remember that the World Health Organisation (WHO) diagnostic classification applied to t-score from DXA are not equivalent to t-score derived from QUS. Nevertheless using validated devices QUS provides a reliable and convenient method for the evaluation of osteoporosis risk (Collinge *et al.,* 2010, Hans *et al.,* 2003, Hans *et al.,* 1996). Studies such as Glüer *et al.,* (2004) have demonstrated that QUS can predict osteoporotic fracture in patients and Khaw *et al.,* (2004) has demonstrated the predictive power of the QUS in identifying hip fracture in men as well as women. This is important as the investigations on osteoporosis among men are not as prolific as for women (Herrera *et al.,* 2012).

Identifying and establishing prevalence of osteoporosis can contribute to clinical treatment plans and considering that investigations among the ID population is relatively small by comparison to the broad general population, studies establishing prevalence will contribute to building a clinical picture of the bone health status of adults with ID in Ireland. With that in mind the aim of this paper is to explore the prevalence of osteopenia and osteoporosis among the participants of the *Intellectual Disability Supplement to the Irish Longitudinal study on Ageing* (IDS-TILDA).

8.2 Methods

8.2.1 Study design and participants

All participants from the IDS-TILDA study (N=708) were invited to participate in a suite of health assessments as part of the second wave of data collection. Ethical approval had been granted for the health assessments as part of the main IDS-TILDA study Wave 2. This was obtained from the University of Dublin, Trinity College Dublin and from all the Service Providers involved in the study (see appendix 10).

The health assessment consisted of eight objective health measures which included quantitative ultrasound of the calcaneus bone measuring bone quality. An accessible protocol and objective measurement process was developed in easy read format. All participants invited to the health assessments received easy read explanatory information prior to attending. On the day of the assessments full explanation was given using accessible material and each participant attending provided written consent (see appendix 10). No measurement was undertaken without participant agreement and to improve participation and inclusion the whole assessment process was underpinned with an emancipatory philosophy (see the *Health Fair Protocol* appendix 8).

As part of the main IDS-TILDA study and prior to the health assessments each participant completed a pre-interview questionnaire and a core questionnaire in the form of a face to face interview through a computer assisted programme for interviewing (CAPI). The information collected included general demographic information as well as questions about self-reported doctor's diagnosis of chronic conditions, lifestyle behaviours, personal and family history of fracture and healthcare utilisation. A detailed description of the IDS-TILDA study can be accessed elsewhere (McCarron *et al.,* 2011, Carroll *et al.,* 2014).

In total 604 participants of the IDS-TILDA study participated in the overall health assessments[7] and of those 575 had QUS complete on one or both feet, see *figure 8.0* for QUS flowchart participation.

[7] The overall health assessments were a suite of 8 objective measurements which the researcher titled *Health Fair* to avoid any negative connotations of being tested.

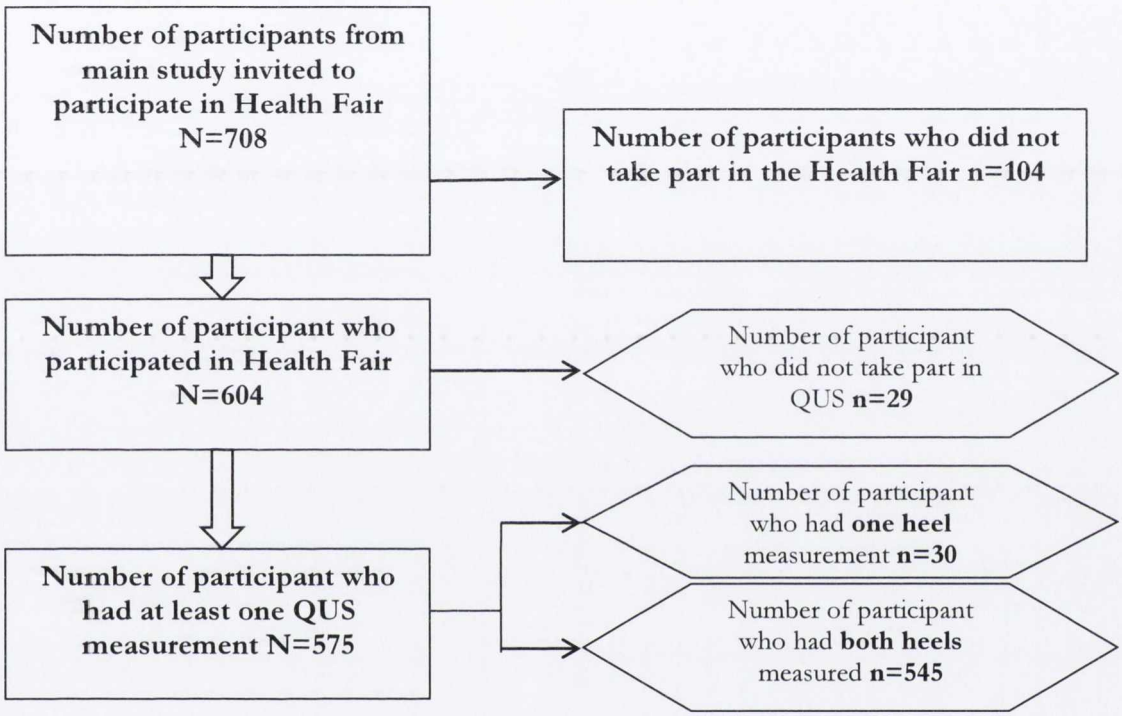


Figure 8.0 Quantitative heel ultrasound participation flowchart

8.2.2 Determining Bone Quality

A GE Lunar Achilles Ultrasonometer was utilised in this study to measure ultrasound variables of the peripheral skeleton namely the os calcis (heel) and a standardised procedure was employed for each participant. All measures were conducted by the same assessor which contributed to the validity of the process. The GE Lunar device is an AquaDry closed self-contained temperature controlled water coupling system contained in silicone membranes. As can be seen in *figure 8.1* water filled membranes hug the heel as ultrasound waves pass from the transmitter to the receiver through the calcaneus bone (Lewiecki *et al.*, 2006). Isopropyl alcohol 70% is used as the coupling agent. The GE Lunar device has built-in reference values based on age and gender of a healthy female Caucasian adult. The references were supplied by the manufacturer and the assessor calibrated the device based on these reference values supplied (young female Caucasian European). The device provides three QUS parameters namely broadband ultrasonic attenuation (BUA in dB/MHz), Speed of Sound (SOS in M/s) and Stiffness Index (SI,%) which the machine calculated and expressed as a QUS t-score. For the purposes of this study the t-score calculated from the stiffness index (SI) identified by the device are divided into three outcome categories and are utilised to classify people into the following categories (Hans *et al.*, 2003). These classifications are utilised for research purposes only and not in a diagnostic capacity.

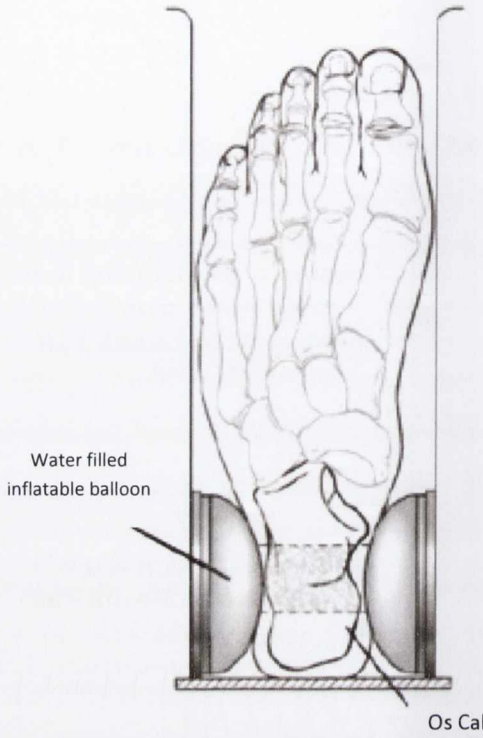


Figure 8.1 Ultrasound passage from transducers (Lewiecki et al., *2006)*

- Normal: QUS t-score > -1.2 (at low risk of fracture)
- Osteopenia: QUS t-score -2.49 ≤ t-score < -1.2 (moderate risk of fracture)
- Osteoporosis t-score≤ -2.5 (at high risk of fracture)

Studies have demonstrated that there can be a difference in QUS results between the left and right foot. The non-dominant foot is recommended to be used for testing however people with ID found it difficult to identify precisely their non-dominant foot, therefore at the time of their QUS assessment both feet were screened where possible. The assessor did instigate measures in an attempt to identify dominance however this could possibly be unreliable in some cases. Also unilateral disrupting factors such as hemiplegia, foot deformity and oedema are more frequently observed among people with ID and therefore could lead to differences between QUS measures of either feet or difficulty obtaining a measure without causing distress. Therefore where obvious contraindications by pathology were noted only one foot measurement was taken. Full easy-read information was presented to the participant, where necessary the participant observed a demonstration of the QUS measurement.

Following instruction and application of the isopropyl alcohol 70% the participant placed their bare foot on the foot plate of the device, the assessor was very conscious to observe for discomfort or anxiety and allay concerns. It was also imperative to ensure the correct positioning of the foot to make certain of an accurate reading. The assessor had pre-prepared the device by inputting the required information, gender, age, which foot, therefore the device was ready for immediate use with no delay. The measurement took 10 seconds in total. A copy of the result was printed and attached to the participants overall results form. The participant received a written easy-read result card (see appendix 12) on completion of their assessments. Any results outside normal parameters were highlighted and the participant was advised to show their results to their carer and doctor.

8.2.3 Quality Assurance

Great care was taken in standardising the method of measurement and ensuring quality control in accordance to the manufacturer's instructions. At the start of each health assessment day the QUS was calibrated and quality assessed using the phantom procedure as advised by the Achilles device. All data collected was entered into an encrypted laptop and all hard copies are stored separate to the consent form in a lock facility in accordance with the Data Protection Act (2003).

8.3 Statistical Analysis and Data Quality

All data was entered into the Statistical Package for Social Science (SPSS) v22 (IBM Corp. 2013) for analysis. A quality checking and a data cleaning procedure was employed. This involved checking five designated identifiers of the total sample, participant PIN, age, height, one blood pressure measure and one t-score, against the hard copy record of each participant. Then a random 10% of all results were checked against the hard copies to identify any anomalies. Descriptive statistics were then undertaken, this involved identifying the mean, median, range and standard deviation to identify outliers, errors and missing values. These were then checked by a second independent assessor. With regards missing values only valid percentages are presented in this paper. The researcher then examined the results of the QUS and the lower of the overall individual scores was chosen as the score of choice to be used for analysis. The scores were then categorised as previously stated. Correlation coefficients were applied to evaluate the relationship between QUS score with demographics, attendance for DXA scan, history of fracture and self-reported doctor's diagnosis of osteoporosis. A result was considered statistically significant if the derived p-value ≤ 0.05.

8.4 Results

As displayed in **table 8.1**, a total of 575 participants had quantitative heel ultrasound (QUS), of those 57.4% (n=330) were female and the majority of participants were between the age of 50 – 64 years (50.8%, n=229). Almost half of the participants had a moderate level of ID (48%, n=252), with 23.3% reporting a mild level and 29% (n=153) reporting a severe/profound level of ID respectively. With regards living circumstances fewer people lived independently or with their family (15.3% n=87), most lived either in community group homes (CGH) (42.7%, n=243) or residential settings (42%, n=239). In total, 14.4% (n=81) reported a self-reported doctor's diagnosis of osteoporosis. Objectively measured QUS identified poorer rates of bone health overall, with over 70% indicating objective evidence of osteopenia (33.2%, n=191) or osteoporosis (41%, n=236). In relation to having DXA screening 18.2% (n=102) reported they had had a DXA in the last 2 years with 7.2% (n=40) stating it was more than 2 years since their DXA scan, however a further 9.7% (n=54) of participants did not know if they had been for DXA but the majority (65%, n=363) reported they had never had a DXA. In total 20.9% (n=116) reported a

history of fracture. People provided information on Accident and Emergency (A&E) attendance in the last year, the greatest reason for attendance was fracture (2.4%, n=13).

Table 8.1: Profile and skeletal health profile of participants who had Quantitative Heel Ultrasound (N=575)

Demographic Profile		*n*	%	Totals*
Gender	Male	245	42.6	575
	Female	330	57.4	
Age	43-49	163	28.3	575
	50-64	229	50.8	
	65+	120	20.9	
Level of ID	Mild	123	23.3	528
	Moderate	252	47.7	
	Severe/Profound	153	29.0	
Living Circumstance	Independent/Family	87	15.3	569
	CGH**	243	42.7	
	Residential	239	42.0	
Skeletal Health Profile				
Self-reported doctor's diagnosis of osteoporosis	Yes	81	14.4	562
Objective Measured QUS	Normal	148	25.7	575
	Osteopenia	191	33.2	
	Osteoporosis	236	41.0	
Attended DXA scan	Yes, within 2 years	102	18.2	559
	Yes, over 2 years	40	7.2	
	No	363	64.9	
	Don't Know	54	9.7	
History of Fracture	Yes	116	20.9	554
	No	438	79.1	
Accident & Emergency attendance	Fracture	13	2.4	552
	Sprain	8	1.4	
	Multiple injuries	2	0.4	

**Not all participants provided information on all questions asked*

As can be seen from **table 8.2** the mean stiffness index was 72.122 (±23.186). With regards QUS t-score the mean score was -2.02 (±1.76) with females scoring lower than males, -2.208 (±1.77) versus -1.78 (±1.734).

Table 8.2: Overall mean score results from the quantitative heel ultrasound N=575

Quantitative Ultrasound	Mean	SD
BUA (dB/MHz)	97.092	±17.937
SOS (m/s)	1522.64	±51.242
Stiffness Index	72.122	±23.186
QUS t-score	-2.026	±1.767
Male QUS t-score	-1.78	±1.734
Female QUS t-score	-2.208	±1.770

The objectively measured bone status was examined stratified by age, level of ID and living circumstance, as seen in **table 8.3**. As would be expected, overall proportionately more females (44.5%, n=147) were identified as having objective evidence of osteoporosis than men, however over a third of the men had QUS objective evidence of both osteoporosis and osteopenia with their levels of osteopenia (34.4%, n=85) proportionately higher than the females (32.1%, n=106). There was a definitive age gradient noticeable within the osteoporosis category with those in the older age group (65years+) demonstrating a greater proportion of objective evidence of osteoporosis (54.2%, n=65). However the same was not true of osteopenia, there was very little difference between the youngest and middle age groups (34.4%, n=56 versus 34.2%, n=100) respectively, both over 5% higher than the older age group (29.2%, n=35). Level of ID appears to have a large impact on objective evidence of osteoporosis with over 6 in 10 of those with severe/profound level of ID presenting with objective evidence of osteoporosis (62.7%, n=96). With regards living circumstance the presence of objective evidence of osteoporosis was very much associated with residential type of living, over half of those in this type of accommodation presented with objective evidence of osteoporosis, 54% (n=129), and a further 31% (n=70) living in these circumstance reporting objective evidence of osteopenia.

Table 8.3: Objectively measured QUS bone status stratified by gender, age, level of ID and living circumstances

Demographic (N=575)	Total	Normal		Osteopenia		Osteoporosis		*p*-value*
		n	**%**	***n***	**%**	***n***	**%**	
Gender								0.114
Male	245	71	29.0	85	34.7	89	36.3	
Female	330	77	23.3	106	32.1	147	44.5	
Age								**0.003**
43 – 49	163	54	33.1	56	34.4	53	32.5	
50 – 64	292	74	25.3	100	34.2	118	40.4	
65+	120	20	16.7	35	29.2	65	54.2	
Level of ID**								**<0.0001**
Mild	123	46	37.4	43	35.0	34	27.6	
Moderate	252	71	28.2	92	36.5	89	35.3	
Severe/Profound	153	18	11.8	39	25.5	96	62.7	
Living Circumstances**								**<0.0001**
Indep/Family	87	40	46.0	25	28.7	22	25.3	
CGH***	243	68	28.0	91	37.4	84	34.6	
Residential	239	36	15.1	70	31.0	129	54.0	

** a=0.05 with statistically significant values in bold*

***Obs missing = Not all participants provided level of ID or living circumstance*

****CGH = Community Group Home*

Table 8.4 presents the profile of those who reported whether or not they attended for DXA (n=142) and those who reported having a history of fracture (n=116). Overall 18% (n=38) of men and 35.4% (n=104) of women reporting having had a DXA scan. The least number of people who attended reported a severe/profound level of ID (20.1%, n=29) compared to those with mild (31.5%, n=34) or moderate level of ID (31.3%, n=68). Living circumstances was not statistically significant with nearly equal proportions of those living independently/family attending as those living in CGH or in residential settings. When considering those whose objective measurement placed them within either the osteopenia or osteoporosis category, almost a quarter of those with objective evidence of osteopenia (23.4%, n=39) and over a third of those with objective evidence of osteoporosis (34.6%, n=72) reported having had a DXA scan. Positively, the majority of those who had a doctor's diagnosis of osteoporosis had had a DXA scan (83.1%, n=64/77).

Of those who had attended for DXA, 35.5% (n=38) reported a prior history of fracture out of the total of 116 participants who reported fracture. This means 67.2% (n=78) of participants with a history of fracture did not attend for DXA, however as the question asked included all history of fracture it is difficult to ascertain if these were specifically osteoporotic type fractures, so therefore these results may be viewed with caution. However, overall hip fracture was one major osteoporotic type established and was reported at 9.5% (n=11) of the total reported fractures. Examining the history of fracture, there were almost equal reported prevalence among males and females 21.7% (n=51) versus 20.4% (n=65) respectively. Age was statistically associated with having experienced a fracture (p-value = 0.03) and also for those who had objective evidence of poor bone health (osteopenia or osteoporosis), p=0.005.

Table 8.4: Profile of those who had a DXA scan and those who reported a history of fracture

Demographic (N=575)	Attended for DXA Scan				Has a history of fracture			
	n	%	No. in sample*	*p*-value	*n*	%	No. in sample*	*p*-value
Gender				**0.0001**				0.705
Male	38	18.0	211		51	21.7	235	
Female	104	35.4	294		65	20.4	319	
Age				**0.0001**				**0.030**
43 – 49	23	16.7	138		23	14.6	157	
50 – 64	65	25.3	257		70	24.9	281	
65+	54	49.1	110		23	19.6	116	
Level of ID*				**0.044**				0.125
Mild	34	31.5	108		33	27.3	121	
Moderate	68	31.3	217		49	20.0	245	
Severe/Profound	29	20.1	144		26	17.4	149	
Living Circumstances*				0.632				0.204
Indep/Family	18	23.7	76		23	26.4	87	
CGH**	62	29.4	211		42	17.8	236	
Residential	62	28.4	218		51	22.1	231	
OM˜ Bone status				**0.025**				**0.005**
Normal	31	23.8	140		21	15.0	140	
Osteopenia	39	23.4	167		32	17.3	185	
Osteoporosis	72	34.6	208		63	27.5	229	
Self-reported doctor's diagnosis of osteoporosis				**0.0001**				0.189
Yes	64	83.1	77		20	26.3	76	
No	77	18.2	422		93	19.7	471	
History of fracture				**0.040**				
Yes	38	35.5	107		-	-	-	-
No	98	25.5	385		-	-	-	
Attended for DXA								**0.040**
Yes	-	-	-		38	27.9	136	
No	-	-	-		69	19.4	356	

*Not all participants provided all data
**CGH=Community Group Home
˜OM=objectively measured
α=0.05

Participants also provided their medication prescriptions. All medicines were recorded verbatim from their prescription charts with the support of their key worker/carer or the field researcher. These were then grouped and coded to identify and classify the medication groups by a pharmacist working on the IDS-TILDA study. Medications that specifically decreased or improved bone health were further grouped into 3 categories by the researcher:

1). Taking at least one medicine that contributed to bone loss

2). Taking at least one treatment medicine for osteoporosis

3). Taking supplementation.

On examination of the bone loss contributing medications, 41% (n=239) were on antiepileptic medicines (AEDs), over a quarter of the sample, 25.4% (n=146), were prescribed proton pump inhibitors (PPI) and a fifth, 20.3% (n=117), were on anti-depressive medicines, selective serotonin reuptake inhibitors (SSRIS). In total, 24.9% (n=143) of participants were on treatment for osteoporosis, most taking a calcium and vitamin D combination 21.4% (n=123), followed by bisphosphates (7.1%, n=41). Of all those who reported their medication use, 4% (n=23) were taking supplementation in the form of vitamin D. Further detail of medicine use can be seen in **table 8.5**.

Table 8.5: Medicines promotion and inhibiting bone loss

Type of Medicine		***n***	***%***
Contributors to bone loss (CBL)	Antiepileptic (AEDs)	239	41.6
	Proton Pump Inhibitors (PPI)	146	25.4
	SSRIS	117	20.3
	Barbiturates	19	3.3
	Lithium	16	2.8
	Oral prednisolone	5	0.9
	Tamoxifen	2	0.3
	Methotrexate Immunosuppressant	2	0.3
	Anastrazole	2	0.3
	Medroxyprogesterone	2	0.3
Taking one or more type of CBL medicine	**Totals**	**368**	**64.0**
Treatment for osteoporosis	Calcium and vitamin D	123	21.4
	Bisphosphates	41	7.1
	Calcium only	15	2.6
	Desosumab	8	1.4
	Teripartide	1	0.2
Taking one or more treatment medicine	**Totals**	**143**	**24.9**
Supplementation	Vitamin D only	23	4.0
	Totals	**23**	**4.0**

SSRIS=Selective Serotonin Reuptake Inhibitors

Medication use was then examined in relation to objective evidence of low bone quality as measured by the QUS, history of fracture and doctor's diagnosis of osteoporosis. In total, 64% (n=368) of those who completed QUS measures were taking medicines that contribute to bone loss. Of those who had objective evidence of osteopenia (n=191), almost two thirds (61.3%, n=117) were on medicines that contribute to bone loss and of those who had objective evidence of osteoporosis (n=236), over three quarters, 77.5%, (n=183) were prescribed medication such as AEDs PPIs or SSRIs. Conversely, for those with objective evidence of osteopenia or osteoporosis, 25.1% (n=48) and 30.1% (n=71) were on treatment for osteoporosis, with a further 5.2% (n=10) and 3.0% (n=7) on vitamin D supplementation respectively.

When examining the medication use in association with history of fracture (n=116), 71.6% (n=83) were prescribed medicines that contribute to bone loss, 29.3%, (n=34) were on treatment for osteoporosis and 3.4%, (n=4) were taking supplementation. Considering those who had a self-reported doctor's diagnosis of osteoporosis (n=81), 80.2% (n=65) were taking medicines that induce bone loss. As would be expected the vast majority, 61 participants (75.3%) were on treatment for osteoporosis. In total for those with a self-reported doctor's diagnosis, 9.9%, (n=8) were taking vitamin D supplement. Full details of medication use in association with QUS results, history of fracture and self-reported doctor's diagnosis of osteoporosis can be seen in **table 8.6**.

Table 8.6: Associations of medications with QUS results, history of fracture and those who self-reported doctor's diagnosis of osteoporosis

Variable*	Medications														Totals
	Contributing to bone loss					Treatment for osteoporosis					Supplementation				
	Yes		No		*p*-value	Yes		No		*p*-value	Yes		No		*p*-value	No. in sample
	n	%	*n*	%		*n*	%	*n*	%		*n*	%	*n*	%		
QUS measures					**0.0001**					0.009					0.492	
Normal	68	45.9	80	54.1		24	16.2	124	83.8		6	4.1	142	95.9		148
Osteopenia	117	61.3	74	38.7		48	25.1	143	74.9		10	5.2	181	94.8		191
Osteoporosis	183	77.5	53	22.5		71	30.1	165	69.9		7	3.0	229	97.0		236
Total	368	64.0	207	36.0		143	24.9	432	75.1		23	4.0	552	96.0		575
History of fracture					0.072					0.198					0.669	
Yes	83	71.6	33	28.4		34	29.3	82	70.7		4	3.4	112	96.6		116
No	274	62.6	164	37.4		103	23.5	335	76.5		19	4.3	419	95.7		438
Total	357	64.0	197	35.6		137	24.7	417	75.3		23	4.2	531	95.8		554
Self-reported doctor's diagnosis of osteoporosis					**0.002**					**<0.0001**					**0.005**	
Yes	65	80.2	16	19.8		61	75.3	20	24.7		8	9.9	73	90.1		81
No	299	62.2	182	37.8		81	16.8	400	83.2		15	3.1	466	96.9		481
Total	364	64.8	198	35.2		142	25.3	420	74.7		23	4.1	539	95.9		562

a=0.05, statistically significant values in bold
**=Not all participants answered all questions*

Consideration was then given to those who have and do not have a diagnosis of osteoporosis. To that end the data was examined with regards those who had reported a doctor's diagnosis of osteoporosis and those who had not, in conjunction with the identified evidence of objectively measured risk of osteopenia and osteoporosis on QUS measurement (see **table 8.7**). More males than females were at a greater risk of presenting with undiagnosed cases of osteoporosis and osteopenia, 93.7%, (n=233) versus 79.6%, (n=258) respectively. Overall, 9 in 10 men (90.8%, n=80) and 7 in 10 women (69.4%, n=100) who presented with objective evidence of osteoporosis risk on QUS did not have a self-reported doctor's diagnosis of osteoporosis. Very few men overall (6.3%, n=15) had a doctor's diagnosis by comparison to women (20.4%, n=66), however the rates of self-reported doctor's diagnosis overall were low (14.4%, n=81). What is of concern is the fact that in total almost 8 in 10 (77.6%, n=180) of all the participants with objective evidence of osteoporosis did not have a doctor's diagnosis.

Table 8.7: Diagnosed and undiagnosed cases of osteoporosis and osteopenia stratified by gender

QUS Objective measurement*		**Self-reported doctor's diagnosis of osteoporosis****						
		Undiagnosed Cases			Diagnosed Cases			Number in sample
		n	**%**	**95% CI**	**n**	**%**	**95% CI**	
Male								
	Normal	63	95.5	87.5 – 98.4	3	4.5	1.6 – 12.5	66
	Osteopenia	80	95.2	83.1 – 95.3	4	4.8	1.8 – 11.0	84
	Osteoporosis	80	90.8	88.4 – 98.1	8	9.1	4.9 – 17.7	88
	Total	223	93.7	89.9 – 96.1	15	6.3	3.9 – 10.1	238
Female								
	Normal	71	93.4	85.5 – 97.2	5	6.6	2.8 – 14.5	76
	Osteopenia	87	83.7	75.4 – 89.4	17	16.3	10.5 – 24.6	104
	Osteoporosis	100	69.4	61.5 – 76.4	44	30.6	23.6 – 38.5	144
	Total	258	79.6	74.9 – 83.7	66	20.4	25.1 – 16.3	324
Total								
	Normal	134	94.4	89.3 – 97.1	8	5.6	2.9 – 10.7	142
	Osteopenia	167	88.8	83.5 – 92.6	21	11.2	7.4 – 16.5	188
	Osteoporosis	180	77.6	75.0 – 85.2	52	22.4	17.5 – 28.2	232
	Total	481	85.6	82.4 – 88.2	81	14.4	11.8 – 17.6	562

**Indicative of risk of the presence of osteoporosis and osteopenia measurement as measured by QUS*
***Missing obs: 12 people who did not answer the question on doctor's diagnosis*
CI= confidence interval

Of interest is the status of those with Down syndrome, following previous investigations in Wave 1 and the indication of poor bone health among this cohort, a brief examination of the DS data was undertaken to examine the differences between diagnosed and undiagnosed cases.

Overall there were 104 participants with DS who participated in the QUS measurement. Well over half, (59.2%, n=61), had evidence of poor bone health, 31.7% (n=33) had objective evidence of osteopenia and 26.9% (n=28) had objective evidence of osteoporosis. **Table 8.8** presents the objective evidence of poor bone health among those with Down syndrome.

Table 8.8 Objectively measured bone quality stratified by type of ID

	Total	Normal		Osteopenia		Osteoporosis		*p*-value*
		n	%	*n*	%	*n*	%	
Type of ID								**0.0001**
Down syndrome	104	43	41.3	33	31.7	28	26.9	
Non-Down syndrome	471	105	22.3	158	33.5	208	44.2	

Similar to the overall study, the results of those with DS were then examined in relation to having a self-reported doctor's diagnosis of osteoporosis. For people with DS, 9 in 10 of those who had objective evidence of osteopenia and almost 7 in 10 of those who had objective evidence of osteoporosis did not have a self-reported doctor's diagnosis of poor bone health. What is of extreme concern is that 100% of the male participants with DS had neither a diagnosis of osteopenia or osteoporosis when comparing results with objective evidence identified. Of the females with DS 83% (over 8 in 10) did not have a diagnosis of osteopenia and 59.1% (almost 6 in 10) did not have a diagnosis of osteoporosis based on the objective evidence identified by QUS (see **table 8.9**).

Table 8.9: Diagnosed and undiagnosed cases of osteoporosis among those with Down syndrome only stratified by gender

QUS Objective measurement*	Self-reported doctor's diagnosis of osteoporosis**						
	Undiagnosed Cases			Diagnosed Cases			
Down syndrome only	**n**	**%**	**95% CI**	**n**	**%**	**95% CI**	No. in sample
Male							
Normal	21	95.5	78.2 – 99.2	1	4.5	0.8 – 21.8	22
Osteopenia	15	100.0	79.6 – 100.0	0	0.0	0 – 20.4	15
Osteoporosis	6	100.0	61.0 – 100.0	0	0.0	0 – 39.3	6
Total	42	97.7	87.9 – 99.6	1	2.3	0.4 – 12.1	43
Female							
Normal	17	85.0	64.0 – 94.8	3	15.0	5.2 – 36.0	20
Osteopenia	15	83.3	60.8 – 94.2	3	16.7	5.8 – 39.2	18
Osteoporosis	13	59.1	38.7 – 76.7	9	40.9	23.3 – 61.3	22
Total	45	75.0	62.8 – 84.2	15	25.0	15.8 – 37.2	60
Totals							
Normal	38	90.5	77.9 – 96.2	4	9.5	3.8 – 22.1	42
Osteopenia	30	90.9	76.4 – 96.9	3	9.1	3.1 – 23.6	33
Osteoporosis	19	67.9	49.3 – 82.1	9	32.1	17.9 – 50.7	28
Total	87	84.5	76.2 – 90.2	16	15.5	9.8 – 23.8	103

**Indicative of risk of the presence of osteoporosis and osteopenia measurement as measured by QUS*
*** Missing obs not all participants answered question on doctor's diagnosis*
CI= confidence interval

8.5 Discussion

Overall 9 in 10 men (90.8%, n=80) and 7 in 10 women (69.4%, n=100) who presented with objective evidence of osteoporosis risk on QUS did not have a self-reported doctor's diagnosis of osteoporosis.

Life expectancy of people with ID has extended and consequently the potential for conditions associated with ageing, such as osteoporosis, is increasing. Osteoporosis is an insidious condition which can have devastating consequences and is correlated directly with ageing. In this present study, over half the population were aged 50 years or over increasing the likelihood of identifying such conditions. Frequently osteoporosis is only diagnosed post an initial clinical fracture, by which time the bone quality is already substantially compromised. For people with ID fracture can contribute further to already established impairment and impact on overall health, quality of life and loss of independence. This present study assessed the bone status of the participants in the longitudinal study on ageing among people with ID (IDS-TILDA). In addition to the objective measurement, participants reported if they had a doctor's diagnosis of osteoporosis, had a history of fracture, had attended for DXA and the medications they were prescribed.

8.5.1 Objectively measured bone health

The IDS-TILDA study identified a self-reported doctor's diagnosis of osteoporosis at Wave 1 of 8.1%, this had doubled to 16.4% by Wave 2 (McCarron *et al.,* 2014), 14.4% of those with self-reported doctor's diagnosis participated in QUS measurement of this current study. Overall, the prevalence of objective evidence of osteopenia and osteoporosis was high with over 74% of participants being identified with poor bone quality. With regards objective evidence of osteoporosis, 41.0% (n=236) of the population in this study presented with a t-score equal to or below -2.5 which confirms findings in previous work by Bastiaanse *et al.,* (2014) who also utilised an Achilles QUS to examine bone health and found 43.7% (n=339) within this category. This differs greatly from the osteoporosis prevalence of 16% (n= 11) identified by Vice *et al.,* (2015) however their study was based on BMD identified through a chart review and the sample was relatively small, they had 69 contributing participants who were based solely in a residential facility, and as can be seen from this study attendance for DXA can be low. In this present study there was no significant difference between gender and the prevalence of low bone health

(objectively measured osteoporosis) at 36.3% males versus 44.5% females, *p*-value=0.114. By comparison in a study carried out in a large residential centre in Holland, Bastiaanse *et al.,* (2014) identified a significant difference in the prevalence of low bone health between males (38.7%) and females (49.5%), *p*-value <0.01.

This study also establishes that age is a significantly associated factor for reduced bone health, *p*-value=0.003, with a definitive age gradient noted for the presence of objective evidence of osteoporosis however, for the presence of osteopenia the association is identified to a greater extent in the younger age groups. Despite this identified gradient and the common assumption and association of osteoporosis with older age, poor bone health in this study is not a condition solely of old age but seen right across all age groups from 43 years onwards. This study is running in tandem with the Irish general ageing study '*The Irish Longitudinal Study on Ageing*' (TILDA), (Barrett *et al.,* 2011). TILDA carried out objective measurement of bone quality in their first wave of data collection and like this study utilised the GE Lunar Achilles QUS. The rates of QUS objectively measured osteoporosis identified in this study are substantially higher than that reported for TILDA whereby they report 3% of males and 13% of females with overall objective evidence of osteoporosis versus 36.3% of males and 44.5% of females in this study. These considerable differences demonstrates the extent of the underlying and unrecognised issue of poor bone health in the Irish population of adults with ID, and what is of great concern the unrecognised high prevalence especially among men. Frequently osteoporosis is seen as a disease that effects women however a report by the International Osteoporosis Foundation (IOF) in an attempt to disparage this myth reported exclusively on men stating that it was time for men to wake up to their risk of osteoporosis. Projections place men on a trajectory of continuous increased risk of fracture in comparison to women which means the message about osteoporosis is not getting through to men (Ebeling 2014). A similar picture is emerging in this study with a more profound message for men with ID and their carer's, osteoporosis is not exclusively a female condition and men with ID along with their carer's need to be concerned.

Men with ID are 12 times more likely to present with objective evidence of osteoporosis than their counterparts in the general population

The association of low bone quality with level of ID was statistically significant, *p*-value <0.0001. Those with severe profound ID were more likely to present with objective

evidence of osteoporosis (62.7%, n=96) and a definitive gradient is also noted as each category is examined, from mild (27.6%, n=34) to moderate (35.3%, n=89). This finding is not surprising considering that those with a severe/profound level of ID are more likely to have mobility issues and greater physical impairment, are more likely to present with polypharmacy and multimorbidity (O'Dwyer *et al.,* 2013, McCarron *et al.,* 2013b). Further, as Wagemans *et al.,* (1998) highlights the issue of immobility contributes greatly to bone status. Considering that bone is a living organ in need of stimulation through movement and impact, and that those with a more severe/profound ID are more likely to be immobile, the implications of lack of exercise in this group as a contributor to poor bone status needs to be highlighted with carer's and support workers. On examination of bone health with regards objective evidence of osteopenia and level of ID, there is very little difference identified between those with a mild or moderate level. This implies that level of ID plays a role in the bone health of the participants in this study. Overall, whatever level of ID the person presents with they are at risk, however it is clear that those with the more severe to profound ID level are at greater risk. There was also a highly significant association between objective evidence of low bone health and living circumstance (p-value<0.0001). Those living in residential type accommodation were more likely to present with evidence of osteoporosis (54%, n=129). This again is not surprising considering that those living in this type of accommodation in Ireland tend to be of an older generation, present with more severe/profound level of ID and have increased health needs (McCarron *et al.,* 2011).

Several studies have identified an association of AED, PPI and SSRIS medications particularly psychotropic medicines with compromised bone integrity (Sato *et al.,* 2001, Targownik *et al.,* 2008, Bolton *et al.,* 2011). These medicines interfere with calcium and vitamin D absorption contributing to bone loss, and the longer the person is taking these medicines or a combination of such medications, the higher the risk. The evidence emerging in this study is that there is a significant association between taking those medicines that contribute to bone loss and objectively measured evidence of osteoporosis and osteopenia (p-value=0.0001), with 77.5%, (N=186) of those on these medicines presenting with QUS evidence of osteoporosis. Whilst two thirds of the participants were on these medications, less than a third with objective evidence of poor bone health were on treatments such as calcichew or bisphosphonates. And, also of concern, is the fact that the vast majority of participants (97%, n=229) with objective evidence of osteoporosis

were not on vitamin D supplementation. Considering that national health policy recommends supplementation, overall supplementation was poor at 4% (n=23). Sunshine levels are poor in Ireland and for those with ID who may infrequently go out in the sun the likelihood of obtaining vitamin D from sunshine is limited. However, Tohill and Laverty (2001) highlight the difficulties and at times impracticality of sun exposure for people with ID, especially those with more severe to profound ID. They suggest that diet plays a more crucial role in these cases, however they note by raising awareness of the importance of vitamin D and incorporating factors such as vitamin D rich diet, and exercise into people's daily care plan, improvement to bone health can be achieved.

8.5.2 Consequence of poor bone health

Fracture occurs as a direct result of bone compromise from trauma and, frequently osteoporotic fracture is as a result of low trauma. Nevertheless as a consequence this will result in a hospital admission which in turn will contribute to distress and pain for the person. Identified fracture rates in this study are high, over one fifth of participants had a history of fracture, however only 27.9% (n=38/116) with a history of fracture attended for diagnostic assessment (DXA). Over 7 in 10 on medication that contribute to bone loss reported a history of fracture and very few people with fracture history were on supplementation (3.4%, n=4). Fracture is the direct result of bone fragility and can lead to reduced quality of life, pain and often surgical intervention. In Ireland it is projected that the number of hospital admissions directly as a result of falls and fractures will almost double if current trends continue, and the impact on an already overburdened health services could possibly be overwhelming (Health Service Executive 2008). There are no specific national figures for people with ID being hospitalised in Ireland, however McCarron *et al.,* (2011) identified 10.4% were hospitalised in the year prior to data collection, with hospital stay ranging from one to twenty days. This is lower than that reported by the national ageing study TILDA at 12.9% (Normand *et al.,* 2011). What is of note is the number of Accident and Emergency (A&E) admissions for people with ID at 18.7% reported by McCarron and colleagues, substantially higher than that reported by TILDA (14.9%). Of even more concern with regards this current study, is the fact that the main reason for A&E attendance was fracture, the devastating clinical outcome of osteoporosis. As noted earlier surprisingly

Men have the highest prevalence of fracture 21.7% versus females at 20.4%

males had the highest prevalence of fracture and it is the males in this study that are least identified.

Globally between 30-40% of all osteoporotic fractures occur among men with a quarter of all hip fractures, the most serious complication of osteoporosis, occurring in men. What is of great concern is that associated mortality is greater among men when compared with women (Bliuc *et al.,* 2009). Osteoporotic fractures account for more disability adjusted life years lost (DALYs) than most common cancers with the exception of lung cancer and among men the life time risk of experiencing an osteoporotic fracture is almost 3 times higher than presenting with prostate cancer (Johnell and Kanis 2006). The implication being is that awareness and education on the management and amelioration of osteoporosis is not reaching its intended audience. Considering the challenges with literacy and education that people with ID experience, and the higher levels of poor bone health identified in this study, the many campaigns through broadcast media and health promotion undertaken by health authorities are certainly not sufficient to get the message to people with ID and their carer's.

8.5.3 Screening and bone status

Clinically diagnosis has become dependent on DXA BMD however what is evident in this study is that the majority of participants did not attend for DXA scanning (65%, n=363), whilst DXA may be contraindicated in some cases, the levels of fracture rate (20.5%, n=116) identified in this study is disproportionately higher than those who had diagnostic screening (64.5% versus 35.5%). Of course it is recognised in the literature that DXA scanning poses challenges for many people with ID, such as the need to travel to a hospital which can contribute to increased anxiety and ultimately difficulty, non-compliance for the assessment and in some cases this results in sedation requirement as well as economical cost implications. However, utilising alternate methods such as the QUS is now proving useful and is recommended in the absence of DXA (Krieg *et al.,* 2008). On examination of DXA attendance it is not surprising that more females (35.4%) than males (18.0%) attended however, what is concerning is that proportionately males in this study had a higher prevalence of fracture rate than females, 21.7% versus 20.4%. It must be remembered that fracture begets fracture and the need for robust skeletal assessment and management especially post initially fracture can stop the fragility fracture cycle.

As would be expected a greater number of the older population attended for DXA, 49.1% (n=54/110) of the over 65 years, however what this means is that over half of the over 65's have never had a DXA scan and even without specific recommendation for people with ID it is recommended that people with one or more risks regardless of age ought to have screening if possible, especially postmenopausal women under 65 years or men aged 70years or older (younger if risk present) (Lin 2012). However, there are no definitive guidelines for screening people with ID although previous studies have made recommendations such as Tyler *et al.,* (2000) who suggests before the age of 50 years, as they note that people with ID ought to be considered a high risk group. This is supported by Dreyfus *et al.,* (2014) who, similar to this current study identified low rates of screening especially among men. Although it must be noted that routine screening in the general population is not recommended, evidence from this study and others (Tyler *et al.,* 2000, Dreyfus *et al.,* 2014) indicate the need to considered targeted purposeful screening. As the aim of screening is to identify risk of fracture and instigate treatment where necessary, further investigation with regards identifying the optimal age at which to screen people with ID in conjunction with identifying a risk matrix, is required. Further, given that screening can contribute to improved health, increased awareness of overall health screening especially bone status screening among service providers is required. Osteoporosis screening for men globally is quite low and the emerging evidence in this study is indicating that this is no different for men with ID. It is most probable that asymptomatic men are less likely to be screened by comparison to asymptomatic women therefore highlighting the issues of screening for men and appropriate identification of when to screen is essential.

8.5.4 Doctors diagnosis versus objectively identified bone status

Self-reported doctor's diagnosis of osteoporosis was reported at 14.4% (n=81). However when this figure is compared with the objective measured evidence of osteoporosis, immense differences are identified. As stated, undiagnosed cases are higher for males, with 9 in 10 males with objective evidence of osteoporosis who did not have a self-reported doctor's diagnosis and 7 in 10 females with objective evidence who did not have a self-reported doctor's diagnosis of osteoporosis. Similar findings are identified when considering osteopenia. Similarly in the general Irish study (TILDA) differences in diagnosed and undiagnosed cases emerged, with men also being identified as those with the predominance for underdiagnoses (Cronin *et al.,* 2011). Overall in this study, the poor

detection of low bone status may be contributed to a number of factors; increased age, early onset of age related conditions, unmet need or difficulty in seeking or expressing health needs, as well as lack of or difficulty in screening, along with the common misnomer of associating osteoporosis specifically as a female condition.

8.5.5 Down syndrome and bone health

Down syndrome has been identified as an independent risk for osteopenia and osteoporosis particularly in males with DS (Angelopoulou *et al.,* 2000, Sakadamis *et al.,* 2002). It this study it has been identified that over half the males with DS who had QUS objective measurement were identified as having either objective evidence of osteopenia or osteoporosis, none of which had a previous diagnosis of osteoporosis. This confirms the findings of these previous studies. Bittles *et al.,* (2007) note that people with DS have an increased predisposition to chronic health conditions such as osteoporosis, sensory impairment, early onset dementia and therefore enhanced mortality. For men with DS issues of premature ageing and early mortality as a consequence of unmet health need, especially the deleterious consequence of osteoporosis must be recognised and considered.

Elsewhere, reports of early mortality of people with ID, that later are identified as unnecessary and preventable (Heslop *et al.,* 2007) continue to occur (Heslop *et al.,* 2014). The consequence of osteoporosis can contribute to death, however osteoporosis is amenable to treatment and prevention, therefore no citizen with or without an ID ought to suffer such a devastating outcome. As people with ID reach middle age their tendency toward obesity and lower levels of physical activity is evident (McCarron *et al* 2011, Burke *et al* 2014), predisposing them to ill health and overall poor health outcomes. With this in mind this present study should prove useful in contributing to establishing the current health picture of adults with ID and providing evaluation for future health need, especially as the life span across the spectrum of ID is increasing. This study also highlights the urgent need for healthcare providers to consider and examine lifestyle behaviours, assessment and screening, and the implications of older age conditions effecting younger cohorts of people with ID as they build plans and strategies for future care and support.

8.6 Limitations

The doctor's diagnosis of osteoporosis is self-report and whilst every effort in this study was made to ensure the accuracy of the data human error is always possible. The bone quality of the participants was established using GE Lunar Achilles QUS, it is acknowledged that this is not the 'gold standard' for measurement. Nevertheless, this chosen method did meet the standards and position laid down by the ISCD. Diagnosis and identification of osteoporosis has moved away from BMD measurement alone. Detection and classification now incorporates a multifaceted approach taking risk factors, lifestyle behaviour and clinical history into account. Another limitation of the study is that fracture data was collected from medical records. Fractures could have occurred that were unrecognised or unreported due to the fact that many people, especially those with a severe/profound level of ID, may have been unable to report. For the purposes of this paper exploring risk factors fell outside the remit and were therefore not considered. However further exploration will be considered in subsequent investigations.

8.7 Conclusion

People with ID are at risk of reduced skeletal integrity. This may possibly be due to a cascade of disparity, poor health behaviours and lack of assessment. The proposed assessment technique used in this study was successful in engaging and establishing the bone health status of 575 participants. However, in consideration to cautions specified by clinical bodies such as ISCD, and numerous studies, QUS measurement alone, lack of standardised protocols and the proliferation of different devices leaves it insufficient for diagnosis. Its usefulness lies in combination with clinical risk factors and as a contribution to instigating prevention. It ought to be recognised that dependence on BMD alone is no guarantee that fracture will not occur, combination of clinical risk factors are required. However on preliminary examination of the common risk factors applied to the general population, for example smoking, excessive alcohol use, parental history, corticosteroid use, their applicability to people with ID is limited. Unless there is a robust method for identifying and monitoring risk factors specific to people with ID and the development of protocols for referring those at risk for further screening, investigation and treatment, osteoporosis and osteopenia will continue to insidiously rise, contributing to increased costs, morbidity and mortality. Therefore there is a need to establish a set of predictors specifically within the field of ID.

The next chapter in this thesis will explore the predictors of osteopenia and osteoporosis in adults with intellectual disability. Drawing from the Wave 2 [2014] data and using the bone status results from the QUS measurements the researcher will explore and examine the significance of all identified risks applicable to people with ID.

CHAPTER 9

PREDICTORS OF POOR BONE HEALTH AMONG OLDER ADULTS WITH AN INTELLECTUAL DISABILITY

"All things are difficult before they are easy."

- John Norley

Chapter Summary

The following chapter explores the predictors for osteopenia and osteoporosis among adults with intellectual disability (ID). The researcher demonstrates how the particular risks were identified and mapped to the data from *the Intellectual Disability Supplement to the Irish Longitudinal Study on Ageing* (IDS-TILDA). The researcher then presents the processes and practices instigated to ensure data quality and integrity. Setting out the variety of approaches available to deal with missing data the researcher highlights the strategy and method employed in this study. The data analysis plan is presented and rationale for employing the two types of prediction modelling strategies is expounded. The profile of the participants is explored and presented. All identified risk factors are descriptively portrayed and inferentially examined in relation to the objective measure of bone status, namely the GE Achilles Quantitative Heel Ultrasound (QUS) categorisation. With consideration to the identified statistically significant predictors the researcher discusses the implications of the findings in light of ID literature. Comparing the logistic regression modelling to the Chi-squared Automatic Interaction Detector Analysis (CHAID) approach the researcher concludes that CHAID is a more user friendly method that would be easily applicable to clinical practice. In conclusion recommendations are made for clinical practice, further research and policy.

9.0 Introduction

Osteoporosis is a term used to describe low bone mineral density (BMD) and microarchitectural deterioration of bone tissue (Kanis *et al.*, 2007). There is no single cause of osteoporosis and BMD is only one component contributing to its diagnosis. There are a variety of contributors from age-related, behavioural lifestyle to epidemiological factors that impact on bone health. Frequently, for people with intellectual disability (ID), the patterns and presentation of morbidity differ greatly from the general population (McCarron *et al.*, 2013a). As people with ID age, their health conditions become more complex and van Schrojenstein Lantman-De *et al.*, (2000) reports a two and half fold increase in conditions among adults with ID in comparison to the general population. Whilst life expectancy for people with ID has greatly increased, earlier onset of older age conditions is evident. However there is a paucity of extensive investigations on many older age conditions with these conditions frequently going unrecognised (Emerson and Hatton 2013, Cooper *et al.*, 2004). One such condition is osteoporosis.

This lack of investigation may be partly due to the difficulties people with ID have in expressing their own health needs (Turner and Robinson 2011). Equally it may be due to the fact that not all conditions are included in health assessments, a fact previously identified by Srikanth *et al.*, (2011) who noted bone health as an area that lacked investigation. Perhaps as many common risk factors such as smoking or alcohol intake, for example, may not factor hugely in the lives of people with ID or there is a lack of information on areas such as parental history. However, considering the insidious nature of osteoporosis it is imperative to investigate what specific risks contribute to bone fragility for people with ID therefore assisting with precise and detailed assessment and diagnosis. With this in mind the aim of this paper is to identify the determinants of poor bone health, namely osteopenia and osteoporosis, among older adults with ID.

9.1 Methods

Data for this paper was obtained from the second wave [2014] of the *Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing* (IDS-TILDA). This included the response information to the pre-interview questionnaire (PIQ) and main face to face interview. This entailed the doctor's diagnosed chronic health conditions, current medications prescribed, behavioural lifestyle (smoking, alcohol use, physical activity), activities of daily living and

parental history of fracture, together with the suite of objective health measures (OM) of Wave 2 [2014]. OM included are height, weight, grip strength, mid-upper arm circumference, waist circumference and quantitative heel ultrasound (QUS).

To increase the reliability of the data and to provide time for participants to seek assistance with its completion, the PIQ, which held the health conditions questions and medicine use, was posted a minimum of one week in advance of the main questionnaire interview. On the day of the face to face interview and on the day of the health assessment, consents were reaffirmed and obtained (in the case of the health assessments) with the participant. Full study protocol detail, design, sampling, participation and ethical approval can be reviewed in Chapter 4 for the IDS-TILDA study and Chapter 7 for the Objective Health Measures.

From extensive general and ID literature searches, the World Health Organisation (WHO) guidelines and the International Osteoporosis Foundation (IOF) guidelines, risk factors were identified and matched to data collected by IDS-TILDA. For reporting clarity and consistency these were categorised into three groups 1) demographics 2) non-modifiable risk factors and 3) modifiable risk factors. Classifications are presented below, see **table 9.1**. Exploration and discussion on clinical risk factors (CRF) associated with people with ID can be reviewed in chapter 3.

Table 9.1 Clinical risk factor framework

Demographics		Non modifiable							Modifiable	
Demographics	**Genetics**	**Musculoskeletal conditions**	**Respiratory**	**Endocrine**	**GIT**	**Neurological**	**Medications**	**Motor function**	**Behavioural Health**	**Medications**
Age	Down syndrome	Scoliosis	Asthma	Hyper-thyroidism	GERD	Stroke/ TIA~	AEDs	Difficulty mobilising 100 yards	Physical activity level	Antipsychotics Barbiturates Lithium
Gender		Rheumatoid Arthritis	COPD	Hypo-thyroidism		Dementia			Smoking	SSIS Corticosteroids
Level of ID		Muscular dystrophy	Lung disease			Spina Bifida		Grip strength	Alcohol consumption > 3units/day	
Living circumstance		Parental history of hip fracture	Cancer✦	Diabetes	Peptic ulceration	Cerebral palsy*			Dietary intake of Ca.	
				Menopause	Cancer✦ Chronic constipation Coeliac disease PKU**				Waist circumference	

**Anoxia at birth effects motor and skeletal development*
~Trans Ischemic Attack
***Phenylketonuria*
✦Cancer is treated as one variable – all reported cancers were collapsed into one variable.
Parathyroidism – no participant reported this condition
Crohns disease – no participant reported this condition
Ulcerative colitis – no participant reported this condition

9.2 Data analysis

9.2.1 Data quality and integrity

All data was analysed using Statistical Package for Social Science (SPSS, Inc., version 22 IBM 2013). As previously described (chapter 8, section 8.3: 172) all the data was scrutinised for anomalies and cleaned. Descriptive statistics and case summaries were conducted to identify valid and missing values.

The principle of Occam's razor states that among several plausible explanations for a phenomenon, the simplest is best. This principle of parsimony was applied as a practical and theoretical approach to choosing variables for the development of the osteopenia and osteoporosis explanatory model (Frost 2015). Unnecessary predictors would add noise to the estimation of other quantities that we are interested in therefore applying this principle to the regression analysis implies that the smallest model that fits the data is best. Due consideration of variables also has the benefit of improving accuracy of the model and ensuring that the chosen variables are theoretically based, avoids '*the fishing expedition*' which is not good practice the consequences being a model that is not applicable to practice interpretation. In fact it can cause an overfitted model which can leave the regression coefficient *p*-values, and R-squared to be misleading and pertain specifically to the researchers' data therefore lacking generalisability. Careful consideration of the variables also assists with addressing collinearity which occurs by having too many variables trying to do the same job. Along with these criteria for choosing variables is the cost element. If the model is to be used for prediction, time and money can be saved by not measuring redundant predictors. Therefore systematic attention was given to the variable selection for the regression modelling and decision tree construction in this study. All variables chosen had a strong theoretical rationale and were initially examined through chi-squared analysis. They were then entered into the multivariate logistic regression models based on their statistical significance in the bivariate analysis. Particular consideration was taken to ensure a balance was found between fitting a model that is too simple and omits important variables and a model that is overfitted, lacks generalisability and is too complex.

9.2.1.1 Addressing missing data

With considerations, valid percentages only are presented in this paper. SPSS version 22 (IBM 2013) offers the options of listwise or pairwise deletion of missing cases or variables where missing values exist. The SPSS options were considered however to do this would affect the study power (Tabachnick *et al.*, 2001). Missing values could have a large effect on predictor variables and cause high case exclusion, therefore the need to explore alternative methods was necessary. Another method frequently used is estimation of missing data through imputation (Duffy 2006). This can consist of median replacement for skewed non-normal distribution (or mean replacement for data normally distributed). The median value is computed from available data and used to replace missing data; this is considered a conservative approach however was deemed inappropriate in this situation. Overall there were 34 candidate variables, 2 continuous and 32 categorical (5 of which are demographic variables). Of the 34 variables 28 (82.35%) were incomplete, in total there were 575 cases, 316 (54.96%) complete and of the values within these cases 97% were complete (see *figure 9.0*).

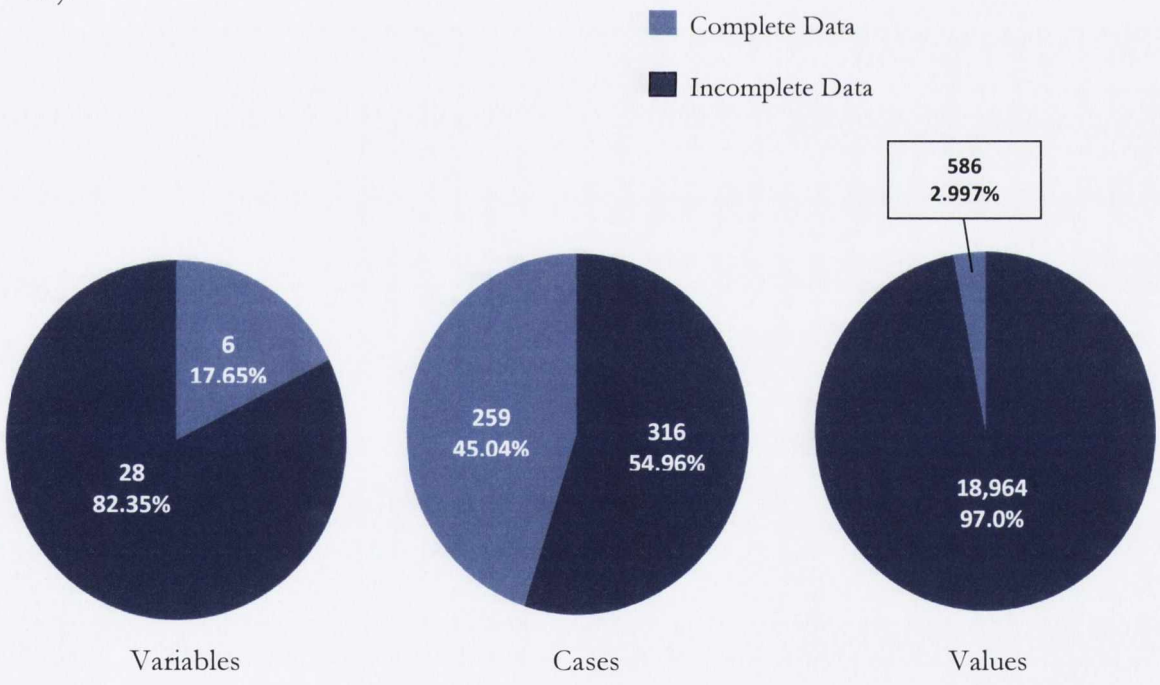


Figure 9.0 Overall summary of missing values

Due to the implications of the missing data, the dataset was scrutinised to identify what missing data existed and if this missing data was missing completely at random (MCAR), missing at random (MAR) or not missing at random (NMAR). Data that are MCAR are difficult to ascertain rationale as the propensity for the data point to be missing is completely random. In contrast the MAR data is when the propensity for the data point to be missing is related to some of the observed values, that is, the data's 'missingness' is conditional of another variable and finally, the NMAR data indicating that it's missingness is related to the data that is missing. To determine the type of missing data, a Missing Values Analysis was conducted in SPSS which can accommodate categorical variables as predictors of missing data. Two variables had over 10% missing (waist classification and grip strength). Following this examination little MCAR was significant therefore data was not missing completely at random A logistic regression model (missing/not missing) was constructed using the 5 demographic variable as predictors. Gender and level of ID are significant predictors of missing waist measurement however as waist measurement was not statistically significant no further action was undertaken with regards this variable. Further examination of the grip strength variable through chi-squared analysis demonstrated level of ID was a significant predictor of missing grip strength values: mild level of ID= 0% missing; moderate level of ID - 11.5% missing and severe/profound level of ID 56.2% missing. Therefore it was deemed necessary to undertake imputation for this variable. To that end the following parametric imputation was conducted.

Parametric imputation is where imputed values for the missing cases are drawn from a parametric distribution which the data is assumed to follow. This distribution was assumed over the grip strength of those with severe/profound level of ID. By examining the spread of grip strengths of those with mild and moderate level of ID, these subsets of the data were found to follow normal distributions therefore those with severe/profound ID were assumed to also follow a normal distribution. In order to parametrically impute the missing values of grip strength, estimates were obtained for the parameters of this distribution through a statistical technique called bootstrapping (Tabachnick *et al.,* 2001, Singh and Xie 2008). Bootstrapping is an alternative to the traditional statistical technique of assuming a particular probability distribution directly from the data. The idea behind bootstrapping is to use the data of a sample study as a "surrogate population", for the purpose of approximating the sampling distribution of a statistic; i.e. to resample (with replacement) from the sample data at hand and create a large number of "phantom

samples" known as bootstrap samples (Singh and Xie 2008). As long as there are more than a few observations the data will reveal their distribution to a reasonable extent. To that end the data was imported into R (R Development Core Team 2011), the variables severe/profound level of ID in relation to grip strength were isolated and thousands of bootstrap samples were generated from the non-missing values. For each bootstrap sample an estimate of the distribution parameters were generated. The mean of these estimates were taken as estimates for the parameters (mean, standard deviation) of the distribution of grip strength for people with severe/profound ID. Imputed values (n=86) were then randomly drawn from the normal distribution with these parameters and used as surrogates for the missing values within the grip strength variable. In order for the imputed values to make physical sense the negative values were set to be equal to zero. One way of describing bootstrapping is that it is sampling from the empirical distribution of the data, letting the data speak for itself and also avoiding overfitting of the model (Tabachnick *et al.*, 2001).

9.2.1.2 Testing for assumptions

With regards interpreting the chi-square for independence all output was checked for violation of the assumptions concerning the minimum expected cell frequency. Those variables that violate the assumption were not included in further analysis. Due to the large number of clinical and behavioural risk factors for osteoporosis and osteopenia, multicollinearity between variables was investigated. Inspection of the correlation matrix for high pairwise correlations was conducted. However since multicollinearity can exist with no pairwise correlations being high, further investigation was performed using SPSS collinearity diagnostics, including Variance Inflation Factor (VIF) and Variance Proportions. The VIF measures the impact of collinearity among the variables in a regression model. The VIF is equivalent to 1/Tolerance. It is always greater than or equal to 1. There is no formal VIF value for determining presence of multicollinearity. Similarly, a tolerance close to 1 means there is little multicollinearity, whereas a value close to 0 suggests that multicollinearity may be a threat (Pallant 2013). The maximum VIF found in this analysis was below 2 (implying the minimum tolerance found was greater than 0.5) indicating multicollinearity is not of concern.

Variance proportions show the percentage of variance of regression coefficient associated with relevant (small) eigenvalue. More than two high values on the sane dimension may be indicative of collinearity. Condition indices of 30 to 100 (generaly indicating moderate to strong collinearities) combined with at least 2 high numbers in a "variance proportion" row is a sign of multicollinearity (Pallant 2013). The final model does not display any signs of multicollinearity.

9.2.2 Identifying sample size

Power analysis is a combination of four elements which are settings for power, alpla, effect size and sample size (Tabachnick *et al.,* 2001, Bannon 2013). Power represents the ability to detect a significant effect if a real difference exists, typically the value is set at 0.80, alpha refers to the level of probability set at 0.05, effect size is set as medium and the sample size is then calculated. These parameters were set for this study. Tabachnick *et al.,* (2001) suggest the following formulae when employing multiple logistic regression for determining sample size.

> $N \geq 50 + 8m$ (where m is the number of independent variables) for testing the multiple correlation, or
>
> $N \geq 104 + m$ for testing individual predictors

(Tabachnick et al., 2001)

The final model contained 16 predictors therefore the sample size required would be 178 to achieve 80% power. For 575 participants in the regression analyses the sample in this study exceeds requirements to achieve power of 80%.

9.2.3 Facilitating data analysis

Reported continuous data were utilised to calculate and categorise, physical activity (IPAQ), grip strength, dietary calcium intake and alcohol intake as follows.

Quantitative Ultrasound (QUS) Categories: For the purposes of this study the t-score calculatd from the stiffness index (SI) identified by the GE Lunar Achilles device is divided into three outcome categories and are utilised to classify people into the following categories (Hans *et al.,* 2003). These classifications are utilised for research purposes only and not in a diagnostic capacity.

1. Normal: QUS T-score $\geq$ -1.2 (at low risk of fracture)
2. Osteopenia: QUS T-score -2.49 $\leq$ t-score < -1.2 (moderate risk of fracture)
3. Osteoporosis < -2.5 (at high risk of fracture)

International Physical Activity Questionnaire (IPAQ): IPAQ was designed to capture physical activity levels across all levels inclusive of leisure-time, work, transportation, and household tasks (Craig *et al.,* 2003). Participants report the frequency and duration of their activity performed for at least a 10 minutes session. The responses were used to categorise the participants into three levels of physical activity based on the standard IPAQ scoring criteria: "low" [physically inactive], "moderate" and "high" levels of physical activity. The moderate level indicates meeting physical activity guidelines of 30 minutes of moderate intensity activity 5 days a week, 20 minutes of vigorous activity 3 days a week, or a combination indicates high levels of physical activity.

Dietary calcium intake: This included total dairy and milk consumption according to portion size, fluid quantity (200ml glass) and frequency (number of times per day/week/ month/not at all) based on the Irish recommendations of the food pyramid (Food Safety Authority 2011). These were then matched according to the recommended portion intake per day and further collapsed into three categories, namely; above recommendations, below recommendations or meets recommended daily allowance.

Alcohol Intake: People were asked to identify the amount and frequency of their alcohol consumption using a modified version of the *Alcohol Use Disorders Identification Test* (Babor *et al.,* 2001). This was then converted to the Food Safety Authority of Ireland recommendations based on the WHO guidelines for hazardous drinking that effects health, stratified by gender, that is; less than 11 units per week for females and less than 17 units for males where a unit measure is equal to half pint of beer or one small glass of wine (Health Service Executive 2012). These were then coded as greater than or equal to ($\geq$) or, less than (<), recommended units of alcohol per week.

9.2.4 Analysis applied

Initial descriptive analyses were conducted to identify prevalence, mean and standard deviation ($\pm$SD) where appropriate. A multistep process was employed to help understand

the contributors to osteopenia and osteoporosis (poor bone health) among adults with ID. The presence of poor bone health is examined through two types of multivariate analysis – logistic regression analysis and decision tree (chi-squared automated interaction detector - CHAID). This has the benefit of enabling comparison across models and best fit. Bivariate analysis (chi-squared) was conducted to determine significant associations between the dependent variable (bone health status) and independent risk factor variables (predictors). Subsequent to this, variables that were significantly related to the dependent variable were then included as predictor variables in the multivariate regression models.

Binary logistic regression was conducted to determine the relative size and magnitude of these bivariate relationships. Two multivariate binary logistic regression models were performed on the categories 1) normal/osteopenia and 2) normal/osteoporosis to identify factors associated with bone status. The demographic variables along with the candidate variables i.e. those statistically significant non-modifiable and modifiable risk factors that emerged from the chi-squared analysis were entered into the model in three blocks. The demographic variables entered as the first block. This block was forced into each model always as the first block and therefore each regression model will always contain the five demographic variables regardless of significance. The non-modifiable risk factors made up the second block and finally the modifiable risk factors made up the third block. Blocks 2 and 3 were entered using the backward likelihood ratio method *Figure 9.1* presents the model with the candidate variables in blocks and their reference categories in parentheses.

Multinomial logistic regression was performed for the final model to identify factors influencing bone status. In this model the dependent variable had three possible outcomes; normal, osteopenia and osteoporosis. The individuals who had a QUS t-score within normal range were the reference group for this analysis. The multinomial regression was conducted using the demographics and the statistically significant candidate variables as previously described with blocks 2 & 3 being entered as one (see *figure 9.1*).

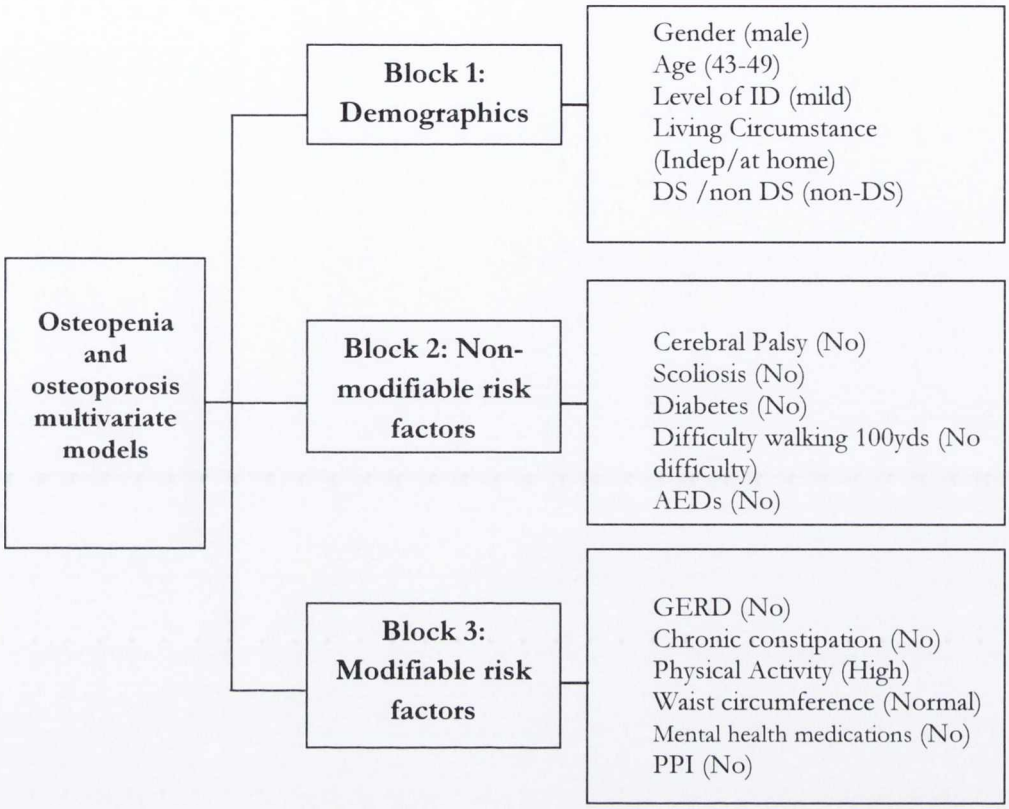


Figure 9.1 Candidate variables & reference in parenthesis for the binary multinomial logistical regression

In addition to the logistic regression models three corresponding models were generated using Chi-squared Automatic Interaction Detector Analysis (CHAID). CHAID allows large data sets of categorical data to be described with one dependent (response) variable and the remainder predictor (explanatory) variables entered into the model. The full list of predictor variables can be seen in **table 9.2**. The CHAID method is a type of decision tree technique which classifies the sample into subgroups to determine the relationship between a dependent and predictor variables using multivariate technique which identifies the size and rank of statistically significant differences to determine the best split based on the logic 'if then' (Miller *et al.,* 2014). In the CHAID analysis, if the dependent variable is nominal, ordinal and continuous, a Chi-squared test, likelihood ratio test and F test respectively is used to specify the best next split at each step. The process is repeated until no other split is identified with consideration to the *p*-value. The method chooses the best partition on the basis of statistical significance and uses the Bonferroni inequality to calculate the significance (Kass 1980, Murphy and Comiskey 2013).

Table 9.2 Predictor variables included in CHAID

Demographics	Health conditions*	Behaviour and family history	Medications
Gender	PKU		Contributing mental health drugs
Level of ID	Coeliac disease	Parental history of hip fracture*	Antiepileptic drugs*
Age	GERD	IPAQ	Proton pump inhibitors
	Peptic ulceration		Corticosteroids
Living circumstance	Hypothyroidism	Grip strength	
	Hyperthyroidism	Smoking	
Menopause*	Asthma	Alcohol intake	
	Lung disease	Waist circumference	
Down syndrome	Spina Bifida	Dietary calcium	
	Alzheimer's/Dementia		
	Muscular dystrophy		
	Stroke/TIA		
	Rheumatoid arthritis		
	Scoliosis		
	Difficulty walking 100 yards		
	Cancer (including leukaemia/lymphoma)		
	Diabetes		
	Cerebral palsy		

**Non-modifiable predictors, apart from demographics all remaining variables were considered modifiable predictors*

There were a number of influencing factors that initiated further examination of the data via CHAID. Firstly, the convenience of CHAID modelling is that all variables without consideration to violating any assumptions can be entered into the model therefore all potential risk factors, which are theoretically and practically sound can be included in the CHAID modelling. Secondly, CHAID modelling can handle a large number of variables and thirdly, the CHAID algorithm has a unique method of handling missing values. No values are discarded instead in the tree growing and classification the user-missing values of nominal independent variables are treated as ordinary values and are included in the analysis as a single combined category. "*For scale and ordinal independent variables the algorithm first generates categories using the valid values and then decides to merge the missing category with its most similar valid category or keep it as a separate category*" (IBM 2011:22).

And finally the fact that all variables are considered the preparation of the data is simpler and the output is easier to interpret making it ideal for clinical use. According to Grady and Berkowitz (2011) who conducted a review on Clinical Prediction Rules (also called clinical decision or prediction models), a good clinical decision rule should be easy

to incorporate into clinical practice, should be based on data from patients for whom the clinical prediction rule will eventually apply and should include all the variables that might be predictive of the clinical outcome. CHAID decision modelling meets all these standards.

9.3 Results

9.3.1 Study profile

In summary, 575 participants had quantitative heel ultrasound measurement. Of those participating 57.6% were female (n=331). The mean age of participants was 56.6years (±9.28). A total of 23.3% (n=123/528) reported a mild level of ID, 47.7% (n=252/528) reported a moderate and 29% (n=153/528) reported a severe/profound level of ID, 8.9% (n=47/528) did not report a level of ID. Those living at home or independently made up the smallest residential grouping of the sample at 15.3% (n=87/569) with almost equal proportions of participants living either in community group homes (CGH) or residential settings, 42% (n=243/569) versus 42.7% (n=239/569). One third of the sample self-reported a doctor's diagnosis of epilepsy with almost half the sample being identified as having severe dependence on the Barthel Index (48.0%, n=272/567). Full demographical profile can be seen in **table 9.3** below.

Table 9.3 Baseline profile of the eligible study participants (N=575)

Variables*		Totals	n	%	Mean ±S.D. (range)
Gender		575			
	Male		244	42.4	
	Female		331	57.6	
Age		575			56.6 ± 9.28 (92-44)
	40 – 49 years		163	28.3	
	50 – 64 years		292	50.8	
	65+ years		120	20.9	
Level of Intellectual Disability		528			
	Mild		123	23.3	
	Moderate		252	47.7	
	Severe/Profound		153	29.0	
Living Circumstances		569			
	Independent/With family		87	15.3	
	Community Group Home		243	42.7	
	Residential Setting		239	42.0	
Type of ID		564			
	Down Syndrome		104	18.4	
	Non Down Syndrome		460	81.6	
	History of fracture	554	116	20.9	
History of falls	In the past month	561	80	14.3	
	In the past year	561	161	28.7	
Epilepsy		566	187	33.0	
Mental Health Conditions		548	284	51.8	
Barthel Index	Independent	567	91	16.0	
	Slight dependence		98	17.3	
	Moderate dependence		106	18.7	
	Severe dependence		272	48.0	
Medications	Antiepileptic medications	575	239	41.6	
	Proton pump inhibitors	575	146	25.4	
Mental health drugs contributing to bone loss		575	143	24.9	

**Not all participants provided responses to all questions asked*

9.3.2 Description of the non-modifiable risk factors for osteopenia and osteoporosis

On examination of the non-modifiable risk factors of those who self-reported arthritis, 2.2%, (n=12/553) reported a doctor's diagnosis of rheumatoid arthritis. Almost three quarters of the females reported having gone through the menopause, the mean age of menopause being 47.6 years (±4.37) for those who reported age at menopause (n=67/292). Mobility difficulties were reported with 34.1% (n=193/566) having difficulty walking 100 yards. In total over two fifths of the sample (41.6%, n=238/575) reported taking antiepileptic medicines. Other conditions and non-modifiable risk factors can be reviewed in **table 9.4**.

Table 9.4 Description of the non-modifiable clinical risk factors identified in study participants (N=575)

Variables*		Totals	n	%	Mean (+SD) max-min
Musculoskeletal	Rheumatoid arthritis (RA)	553	12	2.2	
	Scoliosis	562	28	5.0	
	Parental history of hip fracture	569	20	3.5	
Neurological Conditions	Stroke and /TIA	575	22	3.8	
	Muscular Dystrophy	562	4	0.7	
	Spina Bifida	562	3	0.5	
	Cerebral Palsy	562	37	6.6	
Respiratory Conditions	Asthma	561	29	5.2	
	Other lung disease	558	13	2.3	
Endocrine Conditions	Hypothyroid Disease	562	81	14.4	
	Hyperthyroid Disease	562	16	2.8	
	Diabetes	560	43	7.7	
	Menopause	292	218	74.7	
	Age at menopause	67	-		47.6 ±4.37(35 – 54)
	Menopause at >45 years		49	73.1	
	Early Menopause ≤45 years		18	26.9	
GIT conditions	Phenylketonuria (PKU)	562	7	1.2	
	Coeliac Disease	562	14	2.5	
All Cancer		565	27	4.8	
Motor Function	Difficulty walking 100 yards	566	193	34.1	
Medications	Antiepileptic drugs (AED)		238	41.6	

**Not all participants provided responses to all questions*

9.3.3 Description of the modifiable risk factors for osteopenia and osteoporosis

Participants reported their current smoking behaviour, with 12.4%, (n=70/564) reporting they currently were or previously had smoked. Alcohol consumption exceeding units per week was almost negligible. Almost half the participants' dietary calcium consumption was below the daily recommended allowance (49.2%, n=283/560). Levels of engagement in physical activity were also low with 72.8% (n=410/563) identified at a low level and overweight/obesity was identified at 67.1% (n=380/566). The majority of participants' grip strength was below normal parameters. Conditions such as chronic constipation were reported at 37.7% (n=212/562) among the participants. Over half the participants were on medications that contribute to bone loss. Full description of modifiable risk factors can be viewed in **table 9.5**.

Table 9.5 Description of modifiable risk factors identified in study participants (N=575)

Variables		Total	*n*	%	Mean (+SD) max-min
Have you ever smoked cigarettes, cigars, pipe or cigarillos daily for 1 year		564	70	12.4	
Alcohol consumption		566	214	37.8	
	≥ recommended units per week~		1	0.5	
	< recommended units per week~		213	99.5	
Dietary calcium intake		560			
	Below recommendations		283	49.2	
	Meets recommendations		67	11.7	
	Above recommendations		210	36.5	
GIT conditions	GERD	562	57	10.1	
	Chronic constipation	562	212	37.7	
	Peptic ulceration	536	26	4.5	
Objectively Measured Weight (Kgs)		473			72.8+15.1(93.2-37.9)
MUAC *(cms)		97			29.9+(43-21)
	Underweight		18	3.2	
	Normal		168	29.7	
	Overweight/Obese		380	67.1	
Waist circumference***					
	Normal		85	14.8	
	Increased risk		85	14.8	
	Substantial increased risk		323	56.2	
Physical Activity Level (IPAQ)		563			
	Low		410	72.8	
	Moderate		140	24.9	
	High		13	2.3	
Grip strength (stratified by gender and age)		457			
	Below		442	93.4	
	Normal		15	3.3	
	Above		15	3.3	
Medications	Proton Pump Inhibitors (PPI)		146	25.4	
	Contributing mental health drugs**		143	24.9	
	Corticosteroids		5	0.9	

*~Alcohol consumption stratified by gender :< 17 units/week for males and < 11 units/week for females *MUAC=Mid-upper arm circumference*

***Contributing to bone loss= Selective Serotonin Reuptake Inhibitors (SSRIS), Barbiturates, Lithium*

**** Using WHO cut-offs those having central obesity are at higher risk of developing cardio metabolic complications*

9.3.4 Relationship of the demographic characteristics with the QUS categories of osteopenia and osteoporosis

Participants' demographic characteristics and the QUS t-scores (which were divided into three groups; normal, osteopenia and osteoporosis) were subjected to a chi-squared analysis. The results revealed that overall four demographical variables were statistically associated with the objectively measured QUS results, age (p-value =0.003), Down syndrome (p-value <0.0001), living circumstance (p-value <0.0001) and level of ID (p-value <0.0001). With regards age and objective measure (OM) osteoporosis there is a significant age gradient observed (see linear-by linear p-value table at appendix 16). Similarly, a gradient is detected for level of ID. Those with more severe/profound level of ID are more likely to present with OM osteoporosis according to QUS. The remaining associations can be seen in **table 9.6**.

Table 9.6 Bivariate associations between demographics and QUS measured osteopenia and osteoporosis (bone health status) (N=575)

Demographic*	**QUS t-score**			X^2	*df*	*p-value***
	Normal *n (%)*	**Osteopenia** *n (%)*	**Osteoporosis** *n (%)*			
Gender				4.752	2	0.093
Male	71 (29.0)	85 (34.7)	89 (36.3)			
Female	77(23.3)	106 (32.1)	148 (44.7)			
Age				**16.015**	**4**	**0.003**
43 – 49 years	54 (33.1)	56 (34.4)	53 (32.5)			
50 – 64 years	74 (25.3)	100 (34.4)	118 (40.4)			
65+ years	20 (16.7)	35 (29.2)	65 (54.2)			
Level of ID				**47.173**	**4**	**0.0001**
Mild	46 (37.4)	43 (35.0)	34 (27.6)			
Moderate	71 (28.2)	92 (36.5)	89 (35.3)			
Severe/Profound	18 (11.8)	39 (25.5)	96 (62.7)			
Living Circumstance				**44.836**	**4**	**0.0001**
Independent/Family	40 (46.0)	25 (28.7)	22 (25.3)			
Community Group Home	68 (28.0)	91 (37.4)	84 (34.6)			
Residential Setting	36 (15.1)	74 (31.0)	129 (54.0)			
Type of ID				**18.267**	**2**	**0.0001**
Down Syndrome	43 (41.3)	33 (31.7)	28 (26.9)			
Non-Down Syndrome	105 (22.3)	158 (33.5)	208 (44.2)			

**Not all participants provided responses for all demographics*
*** p-value=0.05, statistically significant results in bold*

9.3.5 Relationship of the modifiable candidate variables with the QUS categories osteopenia and osteoporosis

Further chi-squared analyses were conducted on the non-modifiable and modifiable risk factors (see **tables 9.7** through to **table 9.10**). In total five non-modifiable risks were highly statistically associated with poor bone health (p-value <0.0001) namely, cerebral palsy, difficulty walking, and being on antiepileptic medication. Scoliosis (p-value =0.003) and diabetes (p-value =0.038) were also found statistically significant.

Table 9.7 Bivariate associations between non-modifiable clinical risks factors (non-modifiable predictor variables) and QUS measured osteopenia and osteoporosis (bone health status) (N=575)

Risk Factor	Y/N	QUS t-score			X^2	*df*	*p-value**
		Normal *n (%)*	Osteopenia *n (%)*	Osteoporosis *n (%)*			
Scoliosis					**11.592**	**2**	**0.003**
	Yes	2 (7.1)	6 (21.4)	20 (71.4)			
	No	140 (26.2)	182 (34.1)	212 (39.7)			
Parental History of Hip Fracture					4.170	2	0.124
	Yes	6 (30.0)	10 (50.0)	4 (20.0)			
	No	138 (25.1)	180 (32.8)	231 (42.1)			
Cerebral Palsy					**16.509**	**2**	**0.0001**
	Yes	5 (13.5)	5(13.5)	27 (73.0)			
	No	138 (26.1)	183 (34.9)	205 (39.0)			
Asthma					4.389	2	0.111
	Yes	11 (37.9)	5 (17.2)	13 (44.8)			
	No	131 (24.6)	182 (34.2)	219 (41.2)			
Stroke &/ TIA					1.675	2	0.433
	Yes	3 (13.6)	9 (40.9)	10 (45.5)			
	No	137 (25.6)	177 (33.0)	222 (41.4)			
Hypothyroidism					0.404	2	0.817
	Yes	19 (23.5)	26 (32.1)	36 (44.4)			
	No	123 (25.6)	162 (33.7)	196 (40.7)			
Diabetes					**6.561**	**2**	**0.038**
	Yes	15 (34.9)	18 (41.9)	10 (23.3)			
	No	126(24.4)	168 (32.5)	223 (43.1)			
Menopause					0.295	2	0.863
	Yes	50 (22.9)	70 (32.1)	98 (45.0)			
	No	19 (25.7)	24 (32.4)	31 (41.9)			
All Cancer					4.413	2	0.110
	Yes	11 (40.7)	5 (18.5)	11 (40.7)			
	No	133 (24.7)	182 (33.8)	223 (41.4)			
Waist circumference**					5.503	4	0.239
	Normal	25(29.4)	25 (29.4)	35 (41.2)			
	Increased risk	19 (22.4)	37 (43.5)	29 (34.1)			
	Substantial increased risk	99 (30.7)	120 (37.2)	104 (32.2)			
Difficulty walking 100 yards					**85.348**	**2**	**0.0001**
	Difficulty	19 (9.8)	44 (22.8)	130 (67.4)			
	No Difficulty	124 (33.2)	145 (38.9)	104 (27.9)			
Antiepileptic Medications (AEDs)		**36 (15.1)**	**78 (32.6)**	**125 (52.3)**	**30.784**	**2**	**0.0001**

**Statistically significant results in bold, p-value=0.05*

*** Using WHO cut-offs those having central obesity are at higher risk of developing cardio metabolic complications*

9.3.6 Examining antiepileptic drug use

Antiepileptic medication (AED) are identified as highly significantly associated with the presence of osteopenia and osteoporosis as measured by QUS (X^2 30.784, p-value<0.0001). Further examination of the AED use was undertaken examining the medications in terms of those taking old types medicines and those taking new type medicines and those taking a combination of both old and new type medications (see **table 9.8**). A description of the medication classification can be viewed beneath **table 9.8**. Of note 35.8% (n=206/239) of those taking AED medicines are prescribed old type medicines, 17.4% (n=100/239) are prescribed the new type AED medicines and 11.7% (n=67/239) are taking a combination of both old and new types.

Table 9.8 AED medication usage (N=575)

AED medicine	n	%
Overall	239	41.6
Old type AED medication*	206	35.8
New type AED**	100	17.4
Combination of both old and new type	62	10.8

**Sodium valproate, carbamazepine, phenytoin, phenobarbitone, clonazepam, clobazam, primidone*

***Lamotrigine, levetericam, topimirate, pregablin, zonisamide, gabapentrin*

9.3.7 Examining the association of antiepileptic drug use and QUS objective measurement

Further exploration of the AED medication was conducted examining the association between the different types in relation to QUS objective measurement. It can be seen in **table 9.9** that overall AED use is significantly associated with objective measurement of bone status (p-value<0.0001). There is also a strong significant association observed between taking old type AEDs and the QUS objective measurement (p-value<0.0001) with over half (51.0%, n=105/206) of those taking old type AED being categorise within the osteoporosis range and almost one third (32.5%, n=67/206) within the osteopenia category. New type AED medications were also strongly significantly associated with QUS measures (p-value<0.0001) with 55% (n=55/100) identified within the osteoporosis category. The combination AED were not statistically significant nevertheless 52.2% (n=35/68) of those on a combination (taking one of both old and new types) were categorised within the osteoporosis range. See **table 9.9** for overall associations.

Table 9.9: Association of AED prescribed medication and objectively measured bone status (N=575)

Type of AED		QUS objective measurement						X^2	*df*	*p*-value	Total
		Normal		Osteopenia		osteoporosis					
		n	%	*n*	%	*n*	%				
Overall AED use								**30.784**	**2**	**<0.0001**	
	Yes	36	15.1	78	32.6	125	52.3				239
	No	112	33.3	113	33.6	111	33.0				336
Old type AED*								**18.389**	**2**	**<0.0001**	
	Yes	34	16.5	67	32.5	105	51.0				206
	No	114	30.9	124	33.6	131	35.5				369
New type AED**								**12.405**	**2**	**0.002**	
	Yes	14	14.0	31	31.0	55	55.0				100
	No	134	28.2	160	33.7	181	38.1				475
Combination of both AED types								4.380	2	0.112	
	Yes	12	17.9	20	29.9	35	52.2				68
	No	136	26.8	171	33.7	201	39.6				507

Statistically significant risks in bold, p-value=0.05

**Sodium valproate, carbamazepine, phenytoin, phenobarbitone, clonazepam, clobazam, primidone*

***Lamotrigine, levetericam, topimirate, pregablin, zonisamide, gabapentrin*

9.3.8 Relationship of the non-modifiable candidate variables with the QUS categories osteopenia and osteoporosis

Of the modifiable conditions five variables were statistically significant, chronic constipation, low levels of physical activity and proton pump inhibitors (p-value <0.0001) along with gastroesophageal reflux (p-value=0.001) and low grip strength (p-value =0.003). See **table 9.10** for full examination of the modifiable risk factors.

Table 9.10 Bivariate associations between modifiable risk factors (modifiable predictors) and osteopenia and osteoporosis (bone health status) (N=575)

Risk Factor Y/N	QUS t-score			X^2	*df*	*p-value**
	Normal *n (%)*	Osteopenia *n (%)*	Osteoporosis *n (%)*			
Have you ever smoked for at least a year				1.158	2	0.560
Yes	17 (24.3)	20 (28.6)	33 (47.1)			
No	126 (25.5)	167(33.8)	201 (40.7)			
Dietary Calcium Intake				4.774	4	0.311
Below recommendations	74 (26.1)	85(30.0)	124 (43.8)			
Meets recommendations	13 (19.4)	28 (41.8)	26 (38.8)			
Above recommendations	56 (26.7)	74 (35.2)	80 (38.1)			
GERD			34 (59.6)	**9.42**	**2**	**0.009**
Yes	12 (21.1)	11 (19.3)				
No	130 (25.7)	177 (35.0)	198 (39.2)			
Chronic Constipation			119 (56.1)	**34.356**	**2**	**0.0001**
Yes	32 (15.1)	61 (28.8)				
No	110 (31.4)	127 (36.3)	113 (32.3)			
Peptic Ulceration				1.973	2	0.373
Yes	6 (23.1)	6 (23.1)	14 (53.8)			
No	136 (25.4)	182 (34.0)	218 (40.7)			
Level of Physical Activity				**29.149**	**2**	**0.0001**
Low	82 (20.0)	135 (32.9)	193 (47.1)			
Moderate/High	61 (39.9)	52 (34.0)	40 (26.1)			
Grip Strength**				**13.817**	**2**	**0.001**
Yes	105 (27.8)	134 (35.4)	139(36.8)			
No	36 (45.6)	29 (36.7)	14 (17.7)			
Mental Health Drugs	28(19.6)	49 (34.3)	66 (46.2)	4.079	2	0.130
Proton Pump Inhibitors (PPIs)	**20 (13.7)**	**38 (26.0)**	**88 (60.3)**	**31.699**	**2**	**0.0001**

**Statistically significant risks in bold, p-value=0.05*

***Grip strength stratified by gender and age according to manufacturer's instructions*

****Using WHO cut-offs those having central obesity are at higher risk of developing cardio metabolic complications*

9.3.9 Binary logistic regression identifying crude odds ratios for the candidate variables with the QUS categories osteopenia and osteoporosis

Binary logistic regression was conducted to examine the magnitude and direction for the risks associated with exposure for osteopenia and subsequently examines the magnitude and direction for the risks associated with exposure for osteoporosis. The strongest predictor for osteopenia was living in a residential setting (OR=3.29, 95% CI 1.73-6.23, *p*-value<0.0001). Age and gender had no statistical significance. There were 3 other demographical predictors and 2 non-modifiable and 3 modifiable predictors that were significantly associated with osteopenia occurrence. A total of 17 variables were significantly associated with osteoporosis occurrence. Those who had difficulty walking were over 8 times more likely to present with osteoporosis (OR=8.15, CI 4.72-14.10, *p*-value<0.0001) and those with severe/profound ID over 7 times more likely (OR=7.21, CI 3.69-14.11, *p*-value<0.0001). The remaining predictor results from models 1 and 2 can be seen in **table 9.11**.

Table 9.11 Binary logistic regression identifying crude odds ratios with demographics, risk factors and QUS t-score measured osteopenia and osteoporosis (bone health status) (N=575)

		Osteopenia			Osteoporosis		
		Crude OR	95% CI	*p*-Value*	Crude OR	95% CI	*p*-Value
Demographics							
Gender:	Female	1.150	0.748-1.769	0.525	**1.551**	**1.022-2.352**	**0.039**
Age				0.275			**0.001**
	50 – 64 years	1.303	0.807-2.105	0.279	**1.625**	**1.008-2.619**	**0.046**
	65+ years	1.687	0.868-3.280	0.123	**3.311**	**1.767-6.207**	**0.0001**
Level of ID				0.060			0.0001
	Moderate	1.386	0.825-2.328	0.217	1.696	0.986-2.916	0.56
	Severe/Profound	**2.318**	**1.155-4.650**	**0.018**	**7.216**	**3.690-14.112**	**0.0001**
Living Circumstance				0.001			0.0001
	CGH**	**2.141**	**1.187-3.864**	**0.011**	**2.246**	**1.220-4.136**	**0.009**
	Residential Setting	**3.289**	**1.736-6.231**	**0.0001**	**6.515**	**3.442-12.332**	**0.0001**
Type of ID	Down Syndrome (DS)	**0.495**	**0.295-0.832**	**0.008**	**0.318**	**0.186-0.541**	**0.0001**
Non modifiable risk factors							
Cerebral Palsy		0.749	0.213-2.637	0.652	**3.609**	**1.357-9.600**	**0.010**
Scoliosis		2.308	0.459-11.607	0.310	**6.604**	**1.520-28.697**	**0.012**
Diabetes		0.900	0.437-1.855	0.775	**0.377**	**0.164-0.863**	**0.021**
Difficulty walking 100 yards		**1.98**	**1.099-3.569**	**0.023**	**8.158**	**4.719-14.103**	**0.0001**
Antiepileptic (AED)		**2.147**	**1.337-3.448**	**0.002**	**3.504**	**2.224-5.518**	**0.0001**
Modifiable risk factors							
Chronic Constipation		**1.651**	**1.003-2.717**	**0.049**	**3.620**	**2.262-5.794**	**0.0001**
GERD		0.673	0.288-1.573	0.361	1.860	0.929-3.725	0.080
BMI	Overweight/Obese	**0.533**	**0.304-0.934**	**0.028**	**0.211**	**0.125-0.358**	**0.0001**
Low level of Physical Activity		**1.931**	**1.218-3.061**	**0.005**	**3.589**	**2.232-5.773**	**0.0001**
Grip Strength (above)				0.579			0.666
	Below	1.433	0.427-4.082	0.560	1.674	0.426-6.066	0.433
	Normal	0.800	0.141-4.534	0.801	1.250	0.221-7.084	0.801
Mental Health Medications		1.479	0.876-2.498	0.143	**1.664**	**1.009-2.743**	**0.046**
Proton Pump Inhibitors (PPI)		1.590	0.881-2.868	0.124	**3.805**	**2.217-6.531**	**0.0001**

**p<0.05 All statistically significant risks in bold*
***CGH=Community Group Home*

9.3.10 Multivariate binary logistic regression of QUS measured osteopenia

Demographic variables and variables identified as statistically significant from the binary logistic regression ($\alpha \leq 0.05$) were utilised for the multivariate binary logistic regression (see **table 9.12**). The two models examined were similar to the binary regression. Osteopenia was set as model 1 and osteoporosis was set as model 2.

The final output for model 1 contained the five demographic variables (of which living circumstances and type of ID were significant) and one significant independent variable (difficulty walking 100 yards). The full model containing all predictors was

statistically significant, $p<0.0001$, indicating that the model was able to distinguish between respondent who did and did not present with QUS measured osteopenia. The model as a whole explained between 10.2 % (Cox and Snell R^2) and 13.7% (Nagelkerke R^2) of the variance and correctly classified 64.8% of cases overall. The overall model correctly classified 81.4% of cases with osteopenia. The strongest predictor of reporting the presence of osteopenia was living in a residential setting, recording an adjusted odds ratio (AOR) of 2.27 (CI 1.059-4.878). This indicates that those living in a residential setting were over twice as likely to present with osteopenia controlling for all other factors in the model. Followed by, in descending order, difficulty walking (AOR 2.139, CI 1.102-4.152), severe/profound level of ID (AOR 2.032, CI 0.89-4.637), Community group home (AOR 1.969, CI 0.987-3.93), moderate level of ID (AOR 1.253, CI 0.706-2.225), 65years+ (AOR 1.051, CI0.478-2.315), 50-64 years (AOR 1.045, CI 0.593-1.84), female gender (AOR 0.904, CI 0.547-1493). What is of interest is that those with Down syndrome in this sample were three times less likely to present with osteopenia.

Table 9.12 Multivariate binary logistic regression of the demographics and risk factors associated with QUS t-score measured osteopenia (N=575)

Model 1~Osteopenia as measured and categorised by QUS t-score			
	***p*-value**	**Adjusted Odds Ratio (AOR)**	**95% confidence interval (CI)**
Demographics			
Gender (male)*			
Female	0.693	0.904	0.547-1493
Age (43-49)	0.987		
50 – 64	0.879	1.045	0.593-1.84
65+	0.901	1.051	0.478-2.315
Level of ID (mild)	0.241		
Moderate	0.441	1.253	0.706-2.225
Severe/Profound	0.092	2.032	0.89-4.637
Living circumstance (Indep)	0.089		
Community Group Home	0.055	1.969	0.987-3.93
Residential	**0.035**	**2.273**	**1.059-4.878**
Type of ID (non-DS)			
Down syndrome (DS)	**0.001**	**0.332**	**0.178-0.619**
***Non-Modifiable Risk Factors* (no)**			
Difficulty walking 100 yards	**0.025**	**2.139**	**1.102-4.152**

~Cox and Snell R^2= 0.102, Nagelkerke R^2= 0.137

~Overall Percentage correct predicted 64.9%

~Osteopenia Percentage correct predicted 81.4%

**Reference category in parenthesis with all statistically significant risks in bold (p<0.05)*

9.3.11 Multivariate binary logistic regression of QUS measured osteoporosis

Osteoporosis was set as model 2 and contained 5 demographic variables (of which level of ID and type of ID were significant) and 3 significant independent predictor variables (difficulty walking 100 yards, being on AED medication and diabetes). The full model containing all predictors was statistically significant, X^2 (11, n=342) = 129.60, p<0.0001, indicating that the model was able to distinguish between respondent who did and did not present with QUS measured osteoporosis. The model as a whole explained between 31.5 % (Cox and Snell R^2) and 43.0% (Nagelkerke R^2) of the variance and correctly classified 77.2% of cases overall. The overall model correctly classified 82.6% cases with osteoporosis. As can be seen in **table 9.13** one demographic and three independent variables made a statistically significant contribution to the model with the strongest predictor indicating that those who had difficulty walking were over five and half times more likely to present with osteoporosis than those without difficulty walking (AOR 5.534 CI 2.875-10.654, p-value<0.0001). Having a severe/profound level of ID increased the chances of presenting with osteoporosis four fold compared to those with mild level of ID (AOR 4.299, CI 1.844-10.025, p-value=0.001). Followed by, in descending order, age 65+ (AOR 2.208, CI 0.961-5.072), residential living (AOR 1.906, CI 0.813-4.470), AED (AOR 1.783, CI 1.002-3.172), community group home living (AOR 1.535, CI 0.695-3.389), moderate level of ID (AOR 1.515, CI 0.788-2.914), age 50-64 (AOR 1.489, CI 0.779-2.846), female gender (AOR 1.225, CI 0.696-2.155).

Again in this model those with DS were almost 4 times less likely to present with osteoporosis than those without DS (AOR 0.252, CI 0.119-0.535). It can also be observed that those with diabetes were over 3 less likely to present with osteoporosis (AOR 0.31, CI 0.107-0.9, p-value=0.031).

Table 9.13: Multivariate binary logistic regression of the factors associated with QUS t-score measured osteoporosis (N=575)

Model 2~Osteoporosis as measured and categorised by QUS t-score			
	***p*-value**	**Adjusted Odds Ratio (AOR)**	**95% confidence interval (CI)**
Demographics			
Gender (male)*			
Female	0.482	1.225	0.696-2.155
Age (43-49)	0.167		
50 – 64	0.228	1.489	0.779-2.846
65+	0.062	2.208	0.962-5.072
Level of ID (mild)	0.003		
Moderate	0.213	1.515	0.788-2.914
Severe/Profound	**0.001**	**4.299**	**1.844-10.025**
Living circumstance (Indep)	0.333		
Community Group Home	0.289	1.535	0.695-3.389
Residential	0.138	1.906	0.813-4.47
Type of ID (non-DS)			
Down syndrome (DDS)	**<0.001**	**0.252**	**0.119-0.535**
***Non-Modifiable Risk Factors* (no)**			
Diabetes	**0.031**	**0.31**	**0.107-0.9**
Difficulty walking 100 yards	**<0.001**	**5.534**	**2.875-10.654**
Antiepileptic (AED)	**0.049**	**1.783**	**1.002-3.172**

*~Cox and Snell R^2= 0.315, Nagelkerke R^2= 0.430 ~Osteoporosis Percentage correct predicted 82.6% ~Percentage correct predicted 77.2% *Reference category in parenthesis with all statistically significant risks in bold (p<0.05)*

9.3.12 Multinomial logistic regression of QUS measured osteopenia and osteoporosis

Results from the multinomial logistical regression are presented in table **9.14**. The model contained 5 demographic (of which living circumstance, level of ID and type of ID were significant) and 3 significant independent variables, (proton pump inhibitors, diabetes and difficulty walking 100 yards) significantly predicting variables. The model as a whole explained between 22.7 % (Cox and Snell R^2) and 31.3% (Nagelkerke R^2) of the variance and correctly classified 55.8% of cases overall; 73.7% for osteoporosis and 43.1% for osteopenia. The results demonstrate consistency across a number of factors discriminating osteopenia and osteoporosis from normal bone health. Statistically significant factors to emerge as influencing the presence of osteoporosis include those with a difficulty walking being 6 times more likely to present with osteoporosis than those without difficulty walking (AOR 6.175, 95% CI 3.273 - 11.65, *p*-value<0.001), those with severe/profound level of ID were 4 times more likely to present with osteoporosis than those with mild level of ID (AOR 4.239, 95% CI 1.859 - 9.668, *p*-value=0.001) and those on proton pump inhibitors (PPI) had a 2-fold increased the risk of osteoporosis than those who were not taking PPIs (AOR 2.166, 95% CI 1.139 - 4.118, *p*-value=0.018). With regard osteopenia those living in a residential setting were two times more likely to present with this condition than those who lived independently or with family (AOR 2.204, 95% CI 1.038-4.678). The remaining predictors can be observed in **table 9.14**.

It is also of note that those with DS were less likely to present with osteoporosis than their non-DS peers (AOR 0.179, 95% CI 0.089 – 0.362, *p*-value<0.001) and those with diabetes were over three times less likely to present with osteoporosis (AOR 0.283, 95% CI 0.102 - 0.781, *p*-value=0.015).

Table 9.14 Multinomial logistic regression of the risk factors for QUS t-score measured osteopenia and osteoporosis

Model 3~Osteopenia and osteoporosis as measured and categorised by QUS t-score~ Reference category=normal QUS t-score						
	Osteopenia			Osteoporosis		
	p-value	Adjusted Odds Ratio (AOR)	95% confidence interval (CI)	*p*-value	Adjusted Odds Ratio (AOR)	95% confidence interval (CI)
Gender (male)*						
Female	0.835	0.949	0.578 - 1.558	0.419	1.245	0.732 - 2.116
Age (43-49)						
50 – 64	0.896	1.038	0.591 - 1.825	0.48	1.246	0.677 - 2.29
65+	0.756	1.13	0.521 - 2.453	0.284	1.555	0.693 - 3.487
Level of ID (mild)						
Moderate	0.346	1.312	0.745 - 2.311	0.409	1.306	0.693 - 2.459
Severe/Profound	0.102	1.96	0.874 - 4.394	**0.001**	**4.239**	**1.859 - 9.668**
Living circumstance (Independent)						
Community Group Home	0.063	1.907	0.966 - 3.767	0.209	1.641	0.757 - 3.556
Residential	**0.04**	**2.204**	**1.038 - 4.678**	0.061	2.197	0.964 - 5.01
Type of ID (Non-DS)						
DS	**0.001**	**0.349**	**0.189 - 0.644**	**<0.001**	**0.179**	**0.089 - 0.362**
Proton Pump (No)						
Yes	0.587	1.198	0.624 - 2.299	**0.018**	**2.166**	**1.139 - 4.118**
Walking 100 yards (No difficulty)						
Difficulty	0.056	1.897	0.985 - 3.653	**<0.001**	**6.175**	**3.273 - 11.65**
Diabetes (No)						
Yes	0.819	0.908	0.399 - 2.067	**0.015**	**0.283**	**0.102 - 0.781**

~Cox and Snell R^2= 0.277, Nagelkerke R^2= 0.313

~Percentage correct predicted overall 55.8%

~Osteoporosis Percentage correct predicted 73.7%

~Osteopenia Percentage correct predicted 43.1%

**Reference category in parenthesis with all statistically significant risks in bold ($p<0.05$)*

9.3.13 Chi squared Automatic Interaction Detector (CHAID)

Three CHAID decision tree analyses for the presence of either 1) osteopenia, 2) osteoporosis, and 3) osteopenia and osteoporosis, were performed. The decision tree algorithm partitioned the data into statistically significant subgroups that were mutually exclusive and exhaustive (IBM 2011).

9.3.13.1 CHAID model of the outcome QUS t-score measured osteopenia

In *figure 9.3* the CHAID decision tree analysis shows a two level CHAID tree with a total of 4 nodes, of which 3 are terminal nodes. Terminal nodes (nodes which do not split any further) are the ends of that particular pathway where the prevalence is equated to the likelihood of presenting with osteopenia. The root node commences the decision tree and contains the dependent variable, osteopenia. It can be seen here that 53.6% (n=191/339) of the sample were objectively measured within this category. The CHAID algorithm then splits the root node into two more categories, taking or not taking AED medicines, these are the parent nodes. The split following this are called the child nodes, the independent variable categories Down syndrome or non-Down syndrome in the CHAID analysis tree are the final nodes in this tree and are the final statistically significant predictors influencing the presence of osteopenia. At this point the CHAID decision tree analysis terminates.

The two major variables influencing this model are; taking antiepileptic medications (p-value=0.001) and the presence of Down syndrome (p-value=0.020), the remaining variables entered into the model, age, gender, level of ID, living circumstances, and the remaining modifiable and non-modifiable predictors did not reach significance for inclusion in the model. This model had an overall classification accuracy of 60.8% with its ability to correctly classify osteopenia at 88.5%. Each node has a graphical representation of the dominant outcome associated with the predictor for that particular node. What can be interpreted from this model is that those on AED medicines have a 68.4% chance of presenting with osteopenia. However, examining the branch of the decision tree for those who are not on AED it is indicated that for those not on AED medicines but do have Down syndrome, they are less likely to have osteopenia (37.3%). By comparison those with aetiology other than Down syndrome and are not on AED medicines are more likely to present with osteopenia (54.8%).

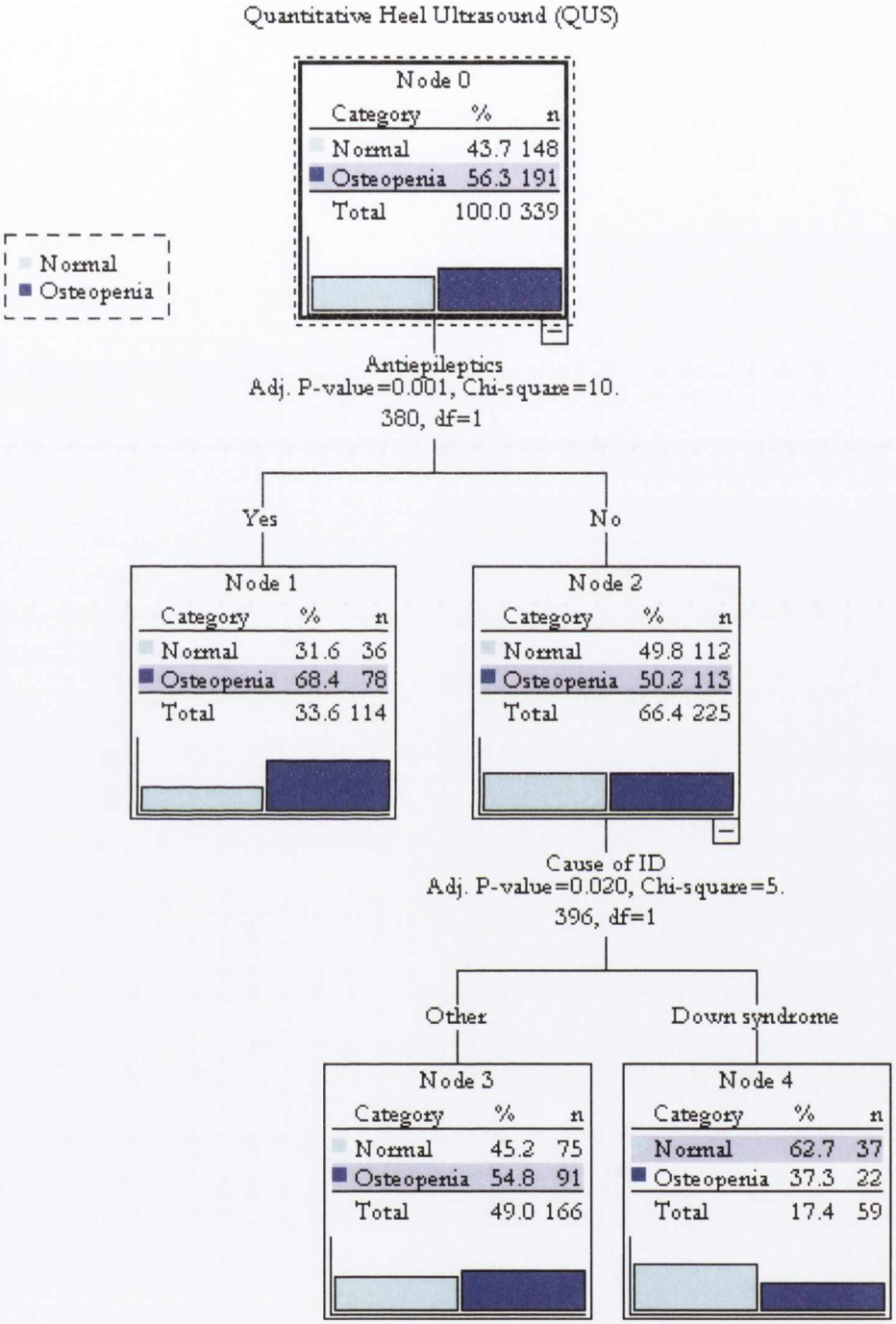


Figure 9.3 Chi-squared automatic interaction detection decision tree analyses of defined predictor variables and the presence of osteopenia

9.3.13.2 CHAID model of the outcome QUS t-score measured osteoporosis

CHAID modelling was again constructed with all predictor candidate variables to identify the most influential variables of those whose QUS t-score placed them within the osteoporosis category. The root node identified that 61.5% (n=236) of participants in this sample were identified within the category osteoporosis. In total, the decision tree partitioned the data initially into 6 nodes with 4 terminal nodes evident. Three major predictor variables reached significance to be included in this model, difficulty walking 100 yards (p-value<0.0001), taking AED medicines (p-value=0.004) and taking proton pump inhibitors [PPIs], (p-value=0.043).

The first level of the tree is split into two branches, difficulty or no difficulty walking 100 yards. It can be seen that those with difficulty walking are more likely to present with osteoporosis (87.2%). This reduced to 82.8% for those not on PPIs (as seen on the final branch of the decision tree) however for those taking PPIs the likelihood of being classified with osteoporosis increases to 93.5%.

Returning to node 1, for those with no difficulty walking they are less likely to present with osteoporosis. CHAID, when dealing with the missing values has also classified the identified missing values within this category. However, for those with no difficulty walking, in the presence of AED medication, the risk increases from 45.1% to 59.4%. This model had an overall classification accuracy of 70.8% with its ability to classify osteoporosis at 72.5%. Just one predictor variable, AED medicines, from the results of this model are similar to the first decision tree *(figure 9.4)* however they are less significant (p-value=0.004) and are placed on the second level of the decision tree. None of the other candidate variables exhibited a significant relationship with the dependent variable.

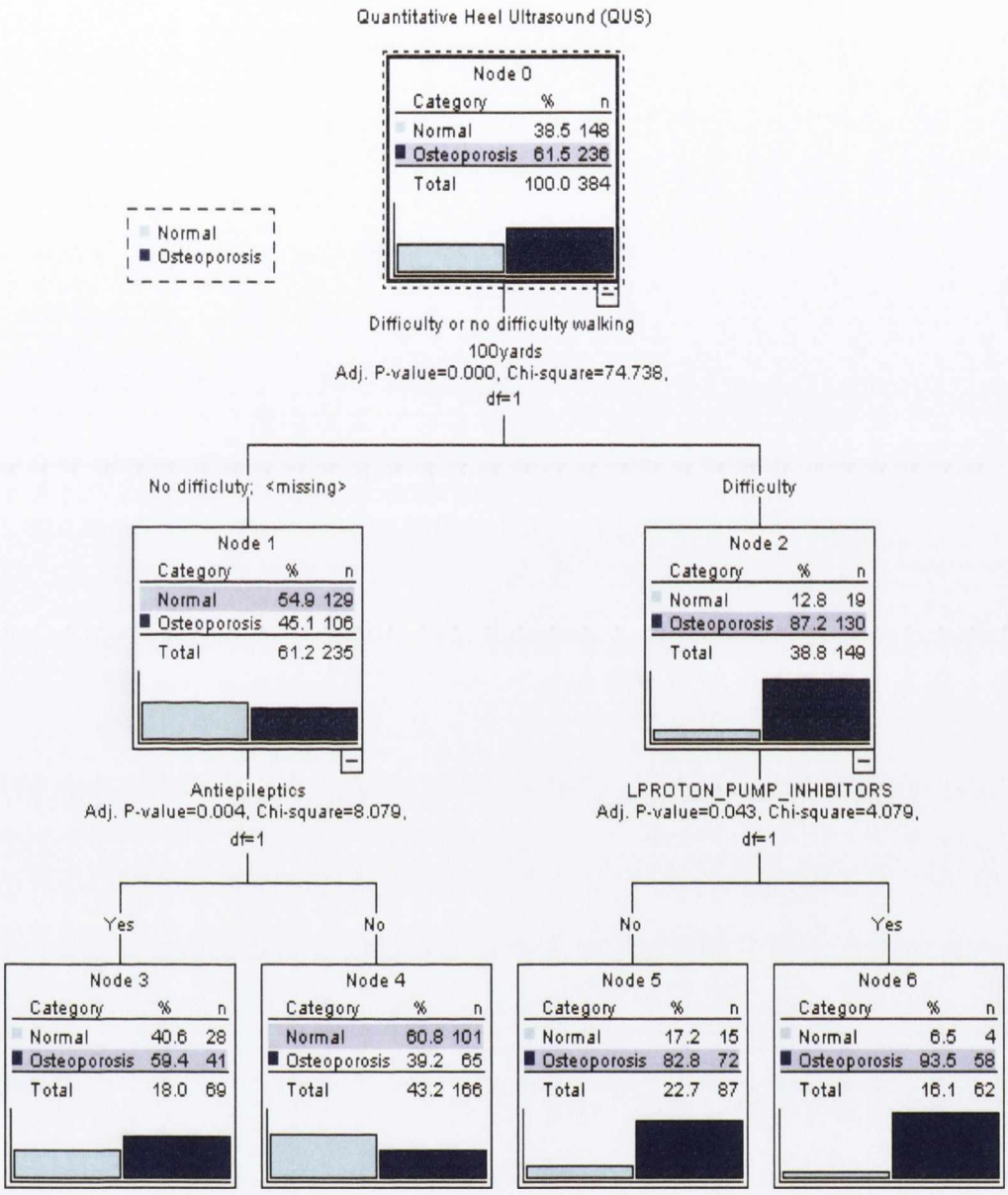


Figure 9.4 Chi-squared automatic interaction detection decision tree analyses of defined predictor variables and the presence of osteoporosis

9.3.13.3 CHAID model of the outcome QUS t-score measured osteopenia and osteoporosis

The tree analysis in *figure 9.5* shows CHAID tree split of three levels with a total of 10 nodes, of which 6 are terminal nodes. Three major predictor variables reached significance to be included in this model including difficulty walking, level of ID and antiepileptic medications. Age, gender, living circumstances, and the remaining modifiable and non-modifiable predictors did not reach significance for inclusion in the model. This model had an overall classification accuracy of 53.0% with its ability to correctly classify osteopenia at 25.1% and osteoporosis at 68.2 %. Each node has a graphical representation of the dominant outcome associated with the predictor for that particular node.

The first level of the tree was split into difficulty or no difficulty walking 100 yards (p-value <0.0001). For those with difficult walking their prevalence rate for osteoporosis was greater than those with no difficulty (67.4% versus 27.7% respectively). Those with no difficulty walking were more likely to present with osteopenia (38.5%) than those within the normal (33.8%) or osteoporosis (27.7%) categories. Examining the second level of the decision tree, level of ID is shown to be the next best predictor of osteopenia and osteoporosis for each of the first level splits. Those with difficulty walking (p-value=0.015) and severe/profound level of ID are more likely to present with osteoporosis than those with difficulty walking with mild/moderate level of ID (79.8% versus 56.7% respectively). Node four terminates with severe/profound level of ID indicating that no further split will occur and that severe/profound level of ID is strongly predictive of osteoporosis. However for those categorised with mild/moderate level of ID the next level indicates that antiepileptic medications (p-value=0.023) is predictive of the likelihood of osteoporosis with those taking AED 66.7% more likely to be classified with osteoporosis than those without AEDs (46%).

Examining the other half of the tree it can be seen that even without any difficulty walking the presence of severe/profound level of ID increases the likelihood of predicting the presence of osteoporosis (43.7%). For those with mild/moderate level of ID with no difficulty walking the risk of osteoporosis decreases and osteopenia is more likely (38.6%). However, in the presence of AED medication the likelihood of osteopenia increases to 49% (p-value<0.0001).

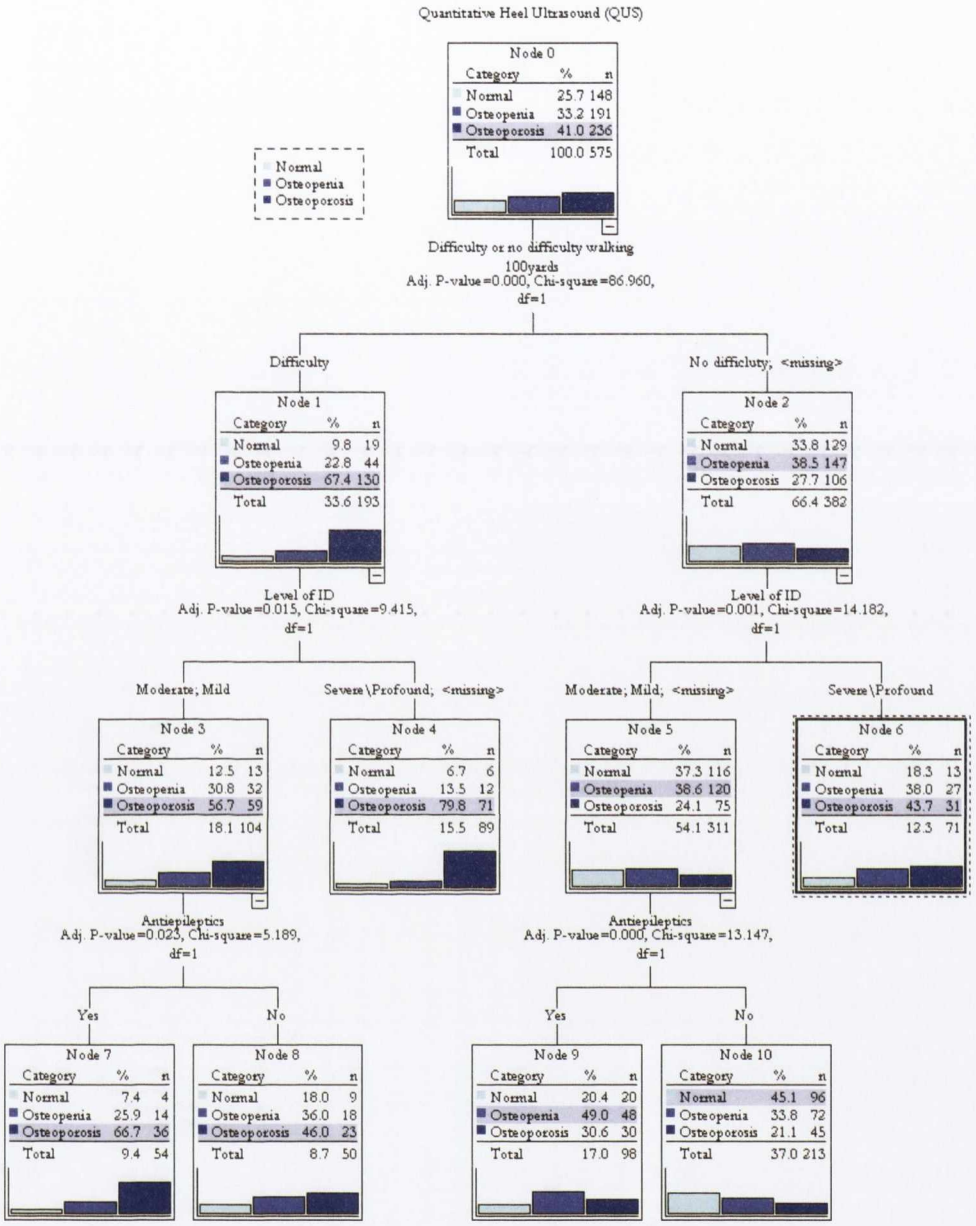


Figure 9.5 Chi-squared automatic interaction detection decision tree analyses of defined predictor variables and the presence of osteopenia and osteoporosis

9.4 Discussion

Commonly identified risk factors observed among the general population such as smoking, alcohol, and corticosteroid use, are not consistently observed at rates to warrant concern.



This study aimed to investigate the contributing factors that increase the risk of poor bone health, namely osteopenia and osteoporosis, among adults with ID. In conjunction with the IDS-TILDA Wave 2 [2014] data, the researcher examined the significant risk factors associated with the QUS results. Risk factors, such as AED use, older age, sex, level of ID and Down syndrome have been identified by a number of ID studies into bone health (Vice *et al.,* 2015, McKelvey *et al.,* 2013, Guijarro *et al.,* 2008, Jaffe *et al.,* 2005, Ray *et al.,* 2002, Angelopoulou *et al.,* 2000, Angelopoulou *et al.,* 1999). Some of these contributors did emerge but by enlarge were not the strongest predictors to emerge from this current investigation.

9.4.1 Emerging issues

This study was conducted among people of all levels of ID living in a variety of settings across Ireland. The results of QUS objective measurement of the heel revealed that 71% (n=174) of the male and 76.6% (n=253) of the female participants had low bone health which demonstrates that the prevalence of poor bone health is high among people with ID irrespective of gender. On review of the regression models, gender did not emerge as a significant predictor. Whilst many studies appropriately focus on women with ID as a high risk category (Arvio *et al.,* 2009, Schrager *et al.,* 2007, Milberger *et al.,* 2002, Jaffe *et al.,* 2001, Foster *et al.,* 2001) only a small number focus on men with ID (see for example Angelopoulou *et al.,* 1999). Considering this, and considering the health and economic cost associated with fracture as a result of osteoporosis, it would be prudent to ensure men with ID had an equal opportunity for screening and investigation as women, as potentially they are at as equal risk as women with ID for compromised bone health.

Some of the stronger predictors to emerge from this study included difficulty walking, severe/profound level of ID, AED medicines and taking PPI medications. The findings with regards mobility issues, level of ID and AED use, support a number of similar studies in the literature (Lin *et al.,* 2015, Bastiaanse *et al.,* 2014, Geijer *et al.,* 2014) who reported high prevalence of osteoporosis among people with ID. Predominantly the

sample sizes are small in comparison to this study with the exception of Bastiaanse *et al.*, (2014) [N=768]. Interestingly evidence of the use of PPIs as a contributor to poor bone health is sparse and tentative among general population studies (Targownik *et al.*, 2008). Following a comprehensive search of the ID literature no studies identifying a relationship between PPIs and bone health were found. Further discussion on this topic follows below.

9.4.1.1 Difficulty walking and level of intellectual disability

The strongest predictor of osteopenia and osteoporosis to emerge from this study was having a difficulty walking (AOR 6.175, 95% CI 3.273 – 11.65, *p*-value<0.001), see table 9.14 and was also highly statistically significant in the CHAID analysis (*p*-value<0.0001). Bone is a living organ in need of stimulation to promote healthy and continuous rejuvenation and bone turnover (Fung 2013). Having difficulty walking not only predisposes the person to an imbalanced bone turnover resulting in increased bone fragility, but also this difficulty can increase the risk of falls and subsequently fracture, the deleterious clinical outcome of osteoporosis. In this situation, difficulty walking contributes further to an ever increasing spiral of risk by making recovery from such events more challenging. People may find it difficult to comply with physiotherapy regimes. Difficulty walking may limit the type and extent of physical intervention that can be engaged in and, difficulty walking increases the complexity of care needed for recovery. In many cases people who have an already pre-existing difficulty walking will not recover fully to attain their pre fracture independence level. As a consequence increasing their dependence and impacting negatively on their quality of life. This emphasises the imperativeness of identifying the bone status of people with ID, ensuring a comprehensive assessment of skeletal status and practical prevention of potential risk. For example, for those with difficulty walking, clear concise assessment of falls risk requires attention and the development of a physical activity program to promote bone health. Further, it is essential that these elements are part of an overall bone health strategy that is incorporated into the person's personal care plans. Through early prevention, fractures and their subsequent health related burden can be significantly reduced.

As previously identified (chapter 8, section 8.4: 172) level of ID was statistically significantly associated with low bone health as identified by QUS measurement with 62.7%, (n=96/153) of those with severe/profound level of ID having poor bone health compared to those with mild level of ID. Considering that those with severe/profound ID

are possibly more likely to be non-ambulant, have higher co-morbidity and have greater dependence and health needs, it is not surprising that this factor emerges as a significant contributing risk factor. Similarly, Schmidt *et al.,* (2004) reports a high prevalence of low BMD among those with a more severe/profound level of ID especially those with complex needs such as immobility, cerebral palsy and/or taking AED medications. Conversely, Zystra *et al.,* (2008) found the differences between mild, moderate, severe and profound were not statistically significant (p-value=0.96) despite noting that the rate of osteoporosis increased as level of ID became more severe. Nevertheless, in this study level of ID is significantly associated with predicting osteopenia and osteoporosis. Whether it is the fact that those with severe/profound level of ID have multiple complex health needs that influence bone health or whether the risk is associated specifically with level of ID, the fact remains that those with severe/profound are at greater risk. This is a group of people who rely on healthcare by proxy therefore due consideration to level of ID is required when assessing the bone health of the person with ID.

9.4.1.2 Antiepileptic medication usage

Over half (51.0%) of those taking old type AED were categorised within the osteoporosis range and almost one third (32.5%) within the osteopenia category.

In Ireland the prevalence of epilepsy is approximately 1% (1 per 100) of those 18 years and older (Linehan *et al.,* 2010). With such a low overall prevalence, it is not surprising that the effects of AED medication on bone health do not get greater attention. It is well documented that AED medication affects bones by interfering with vitamin D metabolism, intestinal absorption of calcium causing calcitonin deficiency and resistance to parathyroid hormone (Babayigit *et al.,* 2006, Mikati *et al.,* 2006). Considering that the prevalence of epilepsy among the cohort in this study was 33% (n=187) and overall 41.6% (n=239) of the participants were taking AED medicines, it is therefore not unexpected that AED medication emerged as a contributory factor. Regardless of all other predictors AED use was identified as doubling the likelihood of osteoporosis for those prescribed this medicine (AOR 1.783, 95% CI 1.002-3.172, p-value=0.049), see table 9.13. Through the CHAID analysis AED usage emerged as one of the most significant contributors to osteopenia (p-value<0.0001).

Examining the type of AED medicines that are prescribed a greater proportion of older type AED medicines are prescribed among this cohort (35.5%, n=206) with a further 17.4%, (n=100) prescribed the new type AED medicines. The newer agents are promised as improved safer agents compared with their older generation AEDs (for example, carbamazepine, and valproate) however for the older generation AED medicines, especially phenytoin, there is an increased effect on BMD which increases the likelihood of bone fragility (Babayigit *et al.,* 2006). The newer second generation AED are reported as having less side effects however the full extent on bone health is still under investigation (French and Gazzola 2011). Increased awareness among carer's and health professionals is required to emphasise the impact AED medications have on bone health. Additionally, for those who are on the older type AED medication, reviews would be prudent and it would be important to ensure skeletal assessment is part of the health care plan for the person with ID. AED medications can leave the bone fragile, coupled with the risk of falling from seizural activity; this increases the vulnerability of the person and leaves them at high risk for skeletal compromise.

9.4.1.2 Proton pump inhibitors and bone integrity

Proton pump inhibitors remain the medicine of choice for upper gastrointestinal conditions and reflux disease. They are potent gastric acid inhibitors and are commonly available as an over the counter medicine. They have strong supporting evidence of their efficacy and safety which probably contributes to a broadly held concern that they are overused (Poulsen *et al.,* 2009). On review of the literature no study evidencing PPI medicines as predictors for osteoporosis among people with ID was found. The dearth of bone health studies among people with ID may be a possible explanation. Nevertheless among the general population evidence is now emerging with regard to the link of PPIs with compromised bone integrity (Corley *et al.,* 2010, Targownik *et al.,* 2008).

The action of PPIs on bone is multifactorial. Firstly, acid inhibition may directly impair calcium absorption, with a resultant decrease in bone density and increase in fracture risk (O'Connell *et al.,* 2005). Secondly, they interfere with magnesium (Mg) dietary absorption, as a result of its need of an acidic environment (Ito and Jensen 2010). Magnesium is a regulator of bone homeostasis. Any reduction in Mg may impair the activity of parathyroid hormones and reduce serum vitamin D (Castiglioni *et al.,* 2013, Matsuzaki *et al.,* 2013). The Corley study identified a significant risk of fracture across all

age groups however noted significance was only present for those with other fracture risks (Corley *et al.,* 2010). They noted age as a determinant of risk. Interestingly those in the younger cohort with one or more risk factors for fracture were at greater risk of bone compromise in the presence of PPIs (e.g. for ages ≥50 years, ≥1 risk factor present OR=1.25, 95% CI 1.16–1.35; risk factors absent OR=0.75, 95% CI 0.43–1.31; for persons <50 years with ≥1 risk factor, OR=1.71, 95% CI 1.00–2.93) (Corley *et al.,* 2010: 5). Of interest in this study is that most of their participants taking PPIs greater than 2 years had at least one other risk factor. In contrast Targownik *et al.,* (2008) found no significant increased risk of bone compromise for those less than 6 years on PPI medication however risk was identified for those taking PPIs more than 6years. This raises the issue of whether length of time on PPI medications is the factor for increasing the risk of bone compromise or the fact that there are other risk factors at play. Further investigation is necessary to examine this issue.

In this current study PPIs emerge as strong contributors to compromised bone integrity. On examination of the data, there was a significant association (p-value<0.0001) with 60% (n=88) of those on PPIs being identified within the osteoporotic category according to the QUS objective measurement compared to those not taking PPI medicines. The initial univariate binary logistic regression model identified a strong significant association between osteoporosis and PPIs (OR 3.805, 95% CI 2.217-6.531, p-value<0.0001). Their effect did not emerge in the multivariate model for osteoporosis. However for the multinomial regression of the combined osteopenia and osteoporosis model, those taking PPIs were identified as twice as likely to present with osteoporosis as those not taking PPIs (AOR 2.166, 95% CI 1.139 - 4.118, p-value=0.018). Further on the CHAID modelling of osteoporosis, PPIs emerge as one of the stronger predictors of osteoporosis (p-value=0.043). CHAID identifies that those taking PPIs with a difficulty walking have a 93.5% risk of being classified with osteoporosis. This brings to mind that many people with severe/profound ID, who are highly likely to have complex needs (Gates 2014), as well as greater risk of multimorbidity (McCarron *et al.,* 2013a) and are perhaps taking multiple medicines (O'Dwyer *et al.,* 2013) may also have concomitant posture and seating compromise, are more susceptible to gastrointestinal conditions such as reflux and are therefore more likely to be taking PPIs. Bone integrity is therefore adversely influenced by this cascade of events highlighting the need for robust education and increased awareness for carer's and staff around the actions and implications of PPIs.

Despite the fact that increase fracture risk as a result of PPI prescription could be confined to those with other risks of fracture, the evidence has raised enough concern for the American Geriatric Society to update the Beers Criteria for inappropriate medicine use among older adults and strongly recommend that older adults should not be prescribed PPIs for a period exceeding 8 weeks unless for high risk patients or specific circumstance, for example oral corticosteroids or erosive esophagitis (Samuel 2015). This implies that it would be prudent to include a comprehensive review of medicines on a regular basis not only for those people with obvious high risk of osteoporosis or fracture, but for all people with ID. Polypharmacy is reported as high among people with ID (O'Dwyer *et al.,* 2013). In light of the easy availability of PPI medicines, for example esomeprazole (Nexium) and the fact that as people with ID move from a more restricted living environment, where medicine use of any preparation was strictly monitored, to more community based living arrangement with more self-control, particularly over the counter preparations, considered safety in medicine practice and awareness is imperative. It should therefore be recommended that carer's and support staff for people with ID be educated in the appropriate use of medicines, especially readily available over the counter preparations. Another issue to highlight with the increasing movement from institutional settings and greater integration into primary health care services is the fact that many primary healthcare professionals would be unfamiliar with people with ID. Therefore education specifically targeted to highlight unique health issues for people with ID would be required.

9.4.2.3 Other concerning factors

An intriguing finding from this study was that those with DS were less likely to present with osteoporosis than their non-DS peers. This is curious considering that a number of studies undertaken among people with ID specifically highlighting that those with DS are at higher risk of osteoporosis (Geijer *et al.,* 2014, Gonzalez-Aguero *et al.,* 2013, McKelvey *et al.,* 2013) and Angelopoulou *et al.,* (2000) going so far as to say that DS is an independent risk factor for osteoporosis. Other issues unique to people with DS provide further support to this theory, such as, the high prevalence of hypogonadism in men (Hsiang *et al.,* 1987), early menopause in women with DS (Schupf *et al.,* 1997), hypotonia and underdeveloped bone growth in adolescence (Hayes and Batshaw 1993), all contribute to a higher risk of bone compromise for those with DS. However, for this study clearer understanding of the results emerges on examination of the CHAID risk pathway. It can

be seen in CHAID that people with DS who are not on AED medication emerge as those who are less likely to present with osteopenia or osteoporosis. It is therefore reasonable to assume that with the absence of epilepsy this cohort are less likely to have Alzheimer's dementia therefore are more likely to be younger, and possibly to be a more healthy active group. Moreover, it is also plausible that other factors were so strong that the effect of the variable DS was hidden. Another possible explanation could be that because of early mortality those with DS were simply not adequately represented. Nevertheless, it is interesting to consider this group and in light of previous descriptive results (Chapter 6: 5) from the Wave 1 [2011] data findings, further examination of osteoporosis and people with DS is warranted.

Also of note is that diabetes emerges as another factor less likely to influence osteopenia or osteoporosis, which is contrary to current literature (Hamann *et al.,* 2012, Lee *et al.,* 2013). A possible explanation for this is the suggestion that some diabetic treatment may be protective (Vestergaard *et al.,* 2005). However for those with type 1 diabetes there is a strong correlation between type 1 diabetes that develops in early adolescence or young adulthood and inadequate accrual of peak bone mass (Hofbauer *et al.,* 2007). It is also suggested that possibly those with type2 diabetes have earlier intervention, including physical exercise programs and dietary monitoring, that promote behavioural lifestyle adjustment, improve health and falls prevention. Despite these posits it is difficult to say without further specific investigation.

9.5 Comparison of the logistic regressions and CHAID models

In this study two prediction methods were applied to estimate the predictive risk factors for the presence of osteopenia and or osteoporosis. The logistic regression with testing for multicollinearity was performed on the risk factors that contribute to poor bone health as a parametric comparison to the CHAID models. The logistic regression gives a better fit, correctly classifying more people and also allows comparison of the strength of the relationship through the odds ratio (OR). The CHAID decision tree allowed multiple variable entries without consideration to assumptions and was an easier method to interpret. The classification of the presence of osteopenia, osteoporosis and a combination of both osteopenia and osteoporosis for both modelling types were not too dissimilar. The predictive variables identified were also similar with the exception of DS and diabetes in the regression modelling. All these variables were reported in previous studies (Matute-

Llorente *et al.*, 2013, Bastiaanse *et al.*, 2014, Mergler *et al.*, 2012, Martínez-Leal *et al.*, 2011, Jaffe *et al.*, 2005). It is reasonable to believe both models are clinically sound. The CHAID compared with the logistic regression model has some advantages. The process of CHAID partitions consecutive data into the best predictive splits. Moreover, given the graphical pathway presentation of the CHAID decision tree it is easier to interpret and easy to use in clinical settings, which supports what Grady and Berkowitz (2011) recommend when they highlight the fact that a predictive model should be easily applied to clinical practice to facilitate appropriate care for high risk patients. This research demonstrated that the CHAID decision tree predictive modelling approach has potential value and can remove the complications of data sets containing a variety of data types (continuous and categorical) as well as missing values to produce clear simple results. In summary, CHAID has the capability of reducing a large dataset to a much smaller number of significant variables and can display complex relationships in a clear and comprehensible way (Murphy and Comiskey 2013).

9.6 Limitations

Despite the valuable findings of this study limitations need to be highlighted. Frequently researchers are challenged by sample size and this study is no different. Future investigations warrant utilising sufficiently larger sample sizes to address sample size issues in an attempt to increase the possibility of findings becoming more generalisable.

Further, all measurements are based on the GE Lunar Achilles QUS heel ultrasound which is not a diagnostic device and therefore may not be reflective of the levels of osteoporosis and osteopenia among this population. However, it is well validated and is a significant predictor of osteoporotic and hip fractures. That said the measurements collected were at one time point only and further measures are required to comprehensively compare results overtime, while we can examine associations, causation cannot be determined.

9.7 Conclusion

The biomechanical structural failure which results in bone fragility is underpinned by a multitude of contributing factors. Overall a number of risk factors were identified in this study. However the strongest contributors among this cohort of adults with ID included difficulty walking, severe/profound level of ID, taking AED medicines and PPI

medications. A number of studies in the literature have previously identified such issues among people with ID. Interestingly, the emergence of PPIs as a predictor is a relatively new issue and currently the researcher could not identify any studies within the ID literature exploring this issue. However, PPI medicines are emerging within the general population as factors that impact on bone health. Their action and how they affect bone has been explored here. In light of this fact, the researcher proposes a number of areas where clinical practice and education would benefit from the findings and proposes that medication reviews are essential to safe and good practice as well as education for those that care and support people with ID.

As people with ID live longer the need to understand the contributing factors for osteoporosis and osteopenia becomes more critical. Considering the unique findings in this study it is important when considering unique presentation of osteoporosis among people with ID to look beyond the 'common' contributing factors. Research into risk factors of osteopenia and osteoporosis among people with ID is sparse and this is especially true for older adults with ID. Much of the studies identified were conducted on small samples which restricts the generalisability of the results. Whilst an advantage of this study is its larger sample the researcher would propose further investigation among a larger ID population sample and perhaps collaboration with investigating groups across Europe. Bone health is essential to successful ageing and for people with ID who require support and assistance to optimise their own health attending to this domain of their lives will contribute to this aim. It will be interesting moving forward as the IDS-TILDA study provides the opportunity for future bone health study, to examine change over time and the impact or influence on health, of undertaking such a work among people with ID.

To that end the next chapter, the concluding chapter of the thesis, provides a comprehensive summary of the overall findings of this thesis, suggests further research endeavours, proposes contributions to educational development and offers potential policy recommendations. Further, a clinical risk assessment algorithm is proposed stemming from the findings of this thesis as a contribution to clinical practice. However further future work is required to test and consolidated such a tool.

CHAPTER 10

BONE HEALTH FOR LIFE

"If you have accomplished all that you have planned for yourself, you have not planned enough."

- Edward Everett Hale

Chapter summary

The final chapter of this thesis presents an overview of the findings of the overall thesis. The findings of each of the studies in the thesis are presented succinctly. From these findings the researcher presents recommendations for clinical practice, further research, for education and policy development and advancement. Noting that this is only the beginning of this work the research identifies the aim of developing these recommendations further into strategies and standards of care delivery. Finally, the researcher presents a provisional bone health armamentarium for clinical practice. Again noting that this is tentative and a vision of what would contribute to clinical practice both within the primary care setting and within the ID field.

10.0 Introduction

This concluding chapter provides an overall summary of the key findings to emerge from this study. Each chapter is presented with a short discussion and the key findings enumerated beneath. From this, key recommendations for research, clinical practice education and policy are extrapolated with suggestions for further and future investigation. In closing this chapter concludes with a potential bone health armamentarium for clinical practice as an initial proposed algorithm that could guide clinical practitioners in assessing the bone health of adults with intellectual disability.

10.1 A systematic review of the prevalence of osteoporosis among adults with an intellectual disability

Peer-reviewed published research was systematically reviewed to identify the prevalence of osteopenia and osteoporosis among adults with intellectual disability. A systematic review protocol was developed to guide the search strategy, assess the quality of each of the included studies and guide data extraction. The researcher was the primary investigator who developed the protocol, conducted the search. In total 50 articles were identified and full text extracted and initially examined. The researcher along with another independent investigator separately scrutinised the articles and applied the study quality assessment criteria. Following this both investigators met and agreed on the final 6 articles going forward for review and data extraction.

Key Findings

- The presentation and reporting of the bone mineral density results varies throughout the reviewed studies. This inconsistency across studies made it impossible to report an overall prevalence for people with ID.

- Along with that fact there is also a proliferation of varying equipment used to examine bone density. No two studies used the same equipment to examine bone density in this review again impact on the ability of the investigator to present overall consensus results.

- Study samples varied, however predominantly sample sizes were small. The average age of participants in the studies examined was 38.76 years, with most studies identifying

their participants with a mild to moderate level of ID. There was also a requirement for mobility in all but one study.

- Overall, people with ID were identified as having lower BMD than their non-ID counterparts with those with Down syndrome reported at greater risk of lower BMD.

- Despite age and activity level there was increased evidence of low BMD among all cohorts implying that osteopenia and osteoporosis among people with ID is not a condition of old age.

- One interesting finding to emerge is that men with ID are likely to be at greater risk of osteoporosis which is of concern as frequently men can be overlooked and osteoporosis traditionally associated as a female condition.

- There is a need for further robust investigation of the bone health of people with ID.

10.2 An exploration of the bone health of older adults with an intellectual disability in Ireland: Results from the first Wave of the IDS-TILDA study.

This paper presented the findings on the bone health of participants in the first wave [2011] of the IDS-TILDA study. The study collected the health, social, community participation and living circumstance data on 753 people aged 40 years and over, across all levels of ID. In an effort to gain precise data a pre-interview questionnaire (PIQ) was posted to people a week in advanced of the face-to-face interview to afford time to seek assistance with gathering the health information from the medical files of the participants. People self-reported their doctor's diagnosis of health conditions, medications, healthcare utilisation and health screening in the PIQ. Among the health conditions self-reported was doctor's diagnosis of osteoporosis.

Key findings

- The prevalence of self-reported doctor's diagnosis of osteoporosis among this cohort was 8.1% (n=61/753) of those, 3% (n=10/338) were males and 12.3% (n=51/415) females.

- People with ID present with many similar risks for osteoporosis as the general population however people with ID have additional risk burden.

- Risk factors among this cohort were high, for example 38.2% (n=287/753) were prescribed antiepileptic medication.

- Over 57% (n=238/415) of the females had experienced the menopause with the mean age of onset at menopause being 47 years, for those who reported their age.

- A large proportion of those interviewed reported overall sedentary lifestyle behaviours with 70.3% (n=528/753) not engaging in exercise at levels that they would accrue health benefit.

- Positive health behaviours to note were that very few participants currently or had ever smoked (15.4%, n=116/753) with very few consuming alcohol above the recommended daily allowance for men and women.

- There was a definitive age gradient observed in cases of self-reported doctor's diagnosis of osteoporosis. Those aged 65 years and over were 6 times more likely to have a diagnosis than their younger cohort (40 – 49 years).

- Females, and people self-reporting a doctor's diagnosis of arthritis, were over four times more likely to have a self-reported doctor's diagnosis of osteoporosis.

- There was a moderately high level of reported history of fractures, 20.6% (n=152/738), the clinical outcome of osteoporosis, and of those who reported a history of fracture, there was very little difference between genders, with males reporting slightly more fractures than females, 20.7% versus 20.5% respectively.

- Of those who reported a diagnosis of epilepsy, 4 in 10 had a history of fracture.

- The highest reported type of fracture among all reported fractures was hip fracture at 12.6% (n=19/152).

- Those who reported difficulty walking were 3 times more likely to self-report a doctor's diagnosis of osteoporosis, with almost 1 in 5 with a doctor's diagnosis of osteoporosis and 3 in 10 reporting a history of fracture.

- Of those who replied to the DXA screening question, 11.1% (n=43/389) had had a DXA scan (Dual energy X-ray Absorptiometry) within the previous 2 years. Such lack of investigation exposes people with ID to an increased risk of fracture.

- Most of the male participants (92.5%, n=161/174), who reported on DXA screening, reported not having had a DXA scan. Similarly, of all those over 65years 81% (n=54/66) reported not having had DXA screening. Interestingly, the most significant factor associated with non-attendance at DXA was difficulty mobilising.

- Overall 17.8% (n=134/753) of participants were taking calcium and vitamin D supplementation, this is despite Irish health recommendations of vitamin D supplementation for all citizens. By comparison, 31.4% of the general population who used food supplements were taking calcium and vitamin D (TILDA).

- Three in every ten participants with a diagnosis of osteoporosis were not prescribed vitamin D, a first line preventative, and over three quarters of those who had a history of fracture were not on vitamin D, calcium or a combination of both.

- Issues to emerge associated with the presence of osteoporosis in this cohort also included eye disease and chronic constipation. Whilst there is no direct correlation between osteoporosis and these two conditions possibly they reflect the more complex health picture of those with ID.

10.3 Osteoporosis and people with Down syndrome: A preliminary descriptive examination of the Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing Wave 1 results.

People with Down syndrome (DS) are recognised as a high 'at risk' group to develop osteoporosis. Among the participants involved in the IDS-TILDA study is a cohort of

people with DS. Their data was extracted from the main sample (n=147/753) and examined in relation to the prevalence of self-reported doctor's diagnosis of osteoporosis, identified risk factors and prevalence of risk factors.

Key findings

- In total 147 people reported they had DS, more female (56%, n=83) than males (44%, n=62). Most participants with DS were below 65 years, with just 3 reporting their age at 65 years all of whom were female.

- A large number of the cohort had no formal education (28.6%, n=42) with just 39.5% (n=58) reporting they attained some primary education.

- In total, 9.7% (n=14/147) reported a history of osteoporosis, all of whom were females, mostly between the ages of 50 – 64 years (27.5%, n=11/40).

- Conditions that increase risk of osteoporosis such are thyroid disease (37.4%, n=55), epilepsy (19.3%, n=28) and arthritis (14.3%, n=21) were evident.

- In total 61% (n=50) of the females reported having experienced the menopause with a further 15.9% uncertain.

- Approximately 13% (n=19) reported having had a fracture, with prevalence twice as high among women (68.4%, n=14) compared to men (31.6%, n=5). Hip fractures were the highest reported type of fracture (26.5%, n=5) experienced.

- With regards behavioural risk, very few reported smoking, consuming alcohol at levels that would impact on bone health however over half (51%, n=76) identified as being relatively inactive.

- Overall, 39.1% (n=25) of men and 34.1% (n=28) of females did not consume the recommended daily requirement of dairy produce.

- Despite the fact that DS is reported as an independent risk for osteoporosis only 8.0% (7/87) reported having had DXA screening.

10.4 Design and development of the health assessment for the Intellectual Disability Supplement to the Irish Longitudinal Study on Ageing

Evidence based guidelines on engaging persons with ID in health assessments and health monitoring is limited. To that end the researcher designed and developed a comprehensive protocol of engaging people with ID in objective health measures. In total eight health assessments were included in the process, namely, grip strength, timed up and go test, waist measurement, hip measurement, height, weight, blood pressure both sitting and standing and finally, quantitative heel ultrasound. To avoid connotations of being measured or tested the researcher called the health assessments '*A Health Fair*' and underpinned the process with the philosophy of emancipation. Thus ensuring the participants were partners in the process, contributed to its development and were afforded every opportunity to engage in the assessments to the best of their ability.

Key findings

- Over 85.3% (n=604/708) of participants in Wave 2 IDS-TILDA [2014] engaged in the *Health Fair*, completing at minimum one objective measure.

- In total, 50.8% (n=307) engaged in all elements of the *Health Fair*. Of those who attended 47.1% were supported to complete their assessments with the assistance of their support staff or carer.

- The mean age of those who attended was 56.4 years. Proportionately the greatest percentage of non-attenders were; male, had a mild level of ID and were within the group whose physical activity level rated low. Further, those with fair or poor self-rated health were over twice more likely not to attend than those with excellent, very good or good self-rated health.

- However, overall there were very little differences in the sociodemographic, behavioural or physical health characteristics of those who attended and those who did not attend the *Health Fair.*

- Some of the contributors to the success of the *Health Fair* included underpinning the process with the fundamentals of emancipation which promoted reasonable adjustment of processes and procedures which ultimately increased success.

- The developments of novel approaches to each of the assessments and so improve participant involvement.

- Carer's support on the day was imperative, contributing to the researchers understanding of the communication style of the participant and encouraging the participant's engagement thus improving the success of the process greatly.

- Acknowledgement of the participants was also imperative and to that end the researcher designed and produced a certificate of achievement, which each participant received on completion of their *Health Fair.*

10.5 Exploring the objectively measured bone status of adults with an intellectual disability, results from the second wave of the IDS-TILDA study

This paper presents the results of the objectively measured bone status of participants in the IDS-TILDA Wave 2 [2014]. Osteoporosis is becoming one of the most common diseases associated with ageing, and is of major public health concern because of the serious consequence. Osteoporosis increases bone fragility thus increasing the risk of fracture, even from low trauma events. Annually osteoporotic fractures costs exceed €59 million in Ireland which does not take into account the social, psychological or personal costs. For people with ID, who frequently present with health challenges and co-morbidities, the risk of undiagnosed osteoporosis is high, thus the risk of subsequent fracture is equally great. In this study the researcher conducted quantitative ultrasound with 575 participants in the *Health Fair.*

- Objectively measured QUS identified poorer rates of bone health overall, with over 70% indicating objective evidence of osteopenia (33.2%, n=191/575) or osteoporosis (41%,

n=236/575). By comparison 14.4% (n=81/562) self-reported a doctor's diagnosis of osteoporosis.

- Combining both osteopenia and osteoporosis QUS objective measured results, 71% (n=174) of the males and 76.6% (n=253) of the females had poor bone health. As would be expected overall proportionately more females (44.5%, n=147/330) were identified as having objective evidence of osteoporosis than men.

- Over a third of the men had QUS objective evidence of both osteoporosis and osteopenia with their levels of osteopenia (34.4%, n=85) proportionately higher than the females (32.1%, n=106).

- Over 6 in 10 of those with severe/profound level of ID presented with QUS objective evidence of osteoporosis (62.7%, n=96).

- Of those who attended for DXA scanning (18.2%, n=102/559) in the last 2 years 18% (n=38) were male and 35.4% (n=64) were female, with those with severe/profound level of ID attending the least (20.1%, n=29/144).

- Over one third of those with objective evidence of osteoporosis reported having had a DXA scan and positively the majority of those who self-reported a doctor's diagnosis of osteoporosis had attended for DXA screening.

- Overall 64% (n=368/575) of participants were taking medications that contribute to bone loss, 41% (n=239/575) were prescribed antiepileptic drugs, over one quarter (25.4%, n=146/575) were on proton pump inhibitors and a fifth (20.3%, n=117/575) were taking antidepressant medicines.

- Of those with QUS objective evidence of osteoporosis over three quarters (77.5%, n=183/236) were taking medicines that contribute to bone loss.

- Overall almost 8 in 10 (77.6%, n=180) of all participants with objective evidence of osteoporosis did not have a self-reported doctor's diagnosis.

- Males are at a greater risk of presenting with undiagnosed cases of osteoporosis and osteopenia, 93.7%, (n=233) versus 79.6%, (n=258) of females respectively. Overall 9 in 10 men (90.8%, n=80) and 7 in 10 women (69.4%, n=100) who presented with objective evidence of osteoporosis risk on QUS did not have a doctor's diagnosis of osteoporosis.

- Men with ID are 12 times more likely to present with objective evidence of osteoporosis than their peers in the general population.

- Of those participants with DS, over half had evidence of poor bone health (59.2%, n=61/103).

- Of the people with DS, 9 in 10 of those who had objective evidence of osteopenia and almost 7 in 10 of those who had objective evidence of osteoporosis did not self-report a doctor's diagnosis of osteoporosis.

- Of extreme concern is that 100% of the male participants with DS had neither a diagnosis of osteopenia, or osteoporosis, when comparing results with QUS objective evidence identified versus self-reported doctor's diagnosis.

10.6 Predictors of poor bone health among older adults with an intellectual disability

This study investigated the contributing risk factors that increase the likelihood of osteopenia and osteoporosis among adults with ID. As people with ID live longer the need to understand the contributing factors to health and particularly bone health is critical. Risk factors were mapped to the data available in the IDS-TILDA Wave 2 [2014] study and examined in relation to the QUS objective measure results.

Key findings

- The data was examined through logistic regression and chi-squared automatic detection (CHAID) decision tree.

- The strongest predictor to emerge was having a difficulty walking. Bone requires stimulation to promote health and continuous rejuvenation; difficulty walking predisposes the person to an imbalance bone turnover contributing to bone fragility. This in turn increases the risk of fracture.

- Level of ID, specifically severe/profound level of ID, was found to be a contributing factor for osteopenia and osteoporosis.

- Proton pump inhibitors (PPIs) emerge as strong contributors for compromised bone integrity. Over 60% (n=88) of those on PPIs were identified within the osteoporotic category according to the QUS measurement and identified as twice as likely to present with osteoporosis as those not taking PPIs.

- The CHAID decision tree classified 93.5% of those who had difficulty walking and also taking PPIs within the osteoporotic category.

- AED medication usage was identified as increasing the risk of osteoporosis almost 2 fold. There are a greater amount of older generation AED preparations utilised among this cohort (34.4%, n=206) with 10.8% (n=62) taking a combination of old and new generation AED medications.

- Over half (51.0%, n=105/206) of those taking old type AED were categorise within the osteoporosis range and almost one third (32.5%, n=67/206) within the osteopenia category.

- Interestingly those with Down syndrome were less likely to present with osteoporosis than their non-DS peers, which is contrary to literature findings. However, on examination of the data through CHAID it emerges that those with DS who are less likely to present with osteoporosis are more likely not to be on AED medicines. This could indicate that they are a cohort who does not have Alzheimer's, a common condition of those with DS with a mean age of 51 years. This could indicate that this cohort is younger and more active.

10.7 Bone health for life

This comprehensive study has identified the paucity of bone health research among people with ID. It has identified a number of areas where improvement can be made and will contribute to the possible development of a specific risk factor predictor model for people with ID. However this is only the foundation work and whilst this thesis encompasses a great deal of work complete it serves only as the beginning. The researcher purports to continue in this area of work and as part of the IDS-TILDA study team has the opportunity to attend to this aim and further investigate areas such as fracture risk. Optimising bone health is a lifelong goal which requires a multifactorial approach. Therefore, stemming from this work the researcher proposes the following clinical practice, research, educational and policy recommendations.

10.7.1 Clinical practice recommendations

A large proportion of people with ID in Ireland were traditionally cared for within institutional type settings provided by services specifically designed for people with ID. Many people living at home and in community group homes also accessed these services for health, day services and respite care. The landscape of care of those with ID is radically changing in Ireland since the publication and adoption of the policy on congregated settings (HSE 2010), whereby all those institutional type settings are expected to close and people living there move to more community integrated settings. Whilst these changes are occurring the shift in the care of people with ID is moving to primary care and more clinical practitioners are being exposed to providing care for people with all levels and complexities of ID. As part of an all-inclusive approach to bone health, comprehensive guidelines specifically designed for people with ID will need to be developed and clinical practice will need to include the following.

I. Comprehensive bone health assessment and clinical guidelines

Bone health assessment should be multidisciplinary and consist of an overall health assessment, assessment of risk factors and behavioural and lifestyle factors. Part of that clinical assessment would include the common risks presented by such algorithms as FRAX® and Q-Fracture® however the additional unique risks identified in this study should also be considered.

- Develop comprehensive bone health guidelines to guide clinical practice in the care and promotion of excellence in bone health of those with ID.

- As part of the comprehensive bone health assessment and clinical guidelines propose the utilisation of the QUS as a feasible, non-stressful easy and quick method of assessing bone quality for people with ID.

- A comprehensive medicine review as part of the overall bone health assessment with consideration to the risks of PPIs and AEDs, and in light of the Beers Criteria for Potentially Inappropriate Medication Use recommendations and being conscious of the prescribing cascade of multiple medicine use.
    - Proton pump inhibitors
    - All antiepileptic drug use
        - Older generation AED (increased risk)

- Identification of specific complex needs of the varying levels of intellectual disability specifically those with severe/profound level of ID.
    - Cerebral palsy
    - Scoliosis
    - Chronic constipation
    - Gastrointestinal reflux disease

- Consideration of other factors that impact on the lives of people with ID such as living circumstance and its impact overtime.
    - Residential living circumstance

- A review of lifestyle behaviours to include physical activity, dietary behaviour and outdoor activity.
    - Activity level and difficulty walking

- Consideration to specific syndromes and the implications on bone health
    - Down syndrome and ID of other aetiologies

- The inclusion of a risk assessment with regards falls. This would include environmental, behavioural and physical ability & physical activity assessment.

- Improved access to specialist such as geriatricians. It is recommended that age limits

on access be removed as people with ID do present with older age conditions at much younger ages, accessing specialities would contribute to improvement in care. To that end and to support specialities such as geriatrics the researcher would propose the development of a clinical nurse specialist in the area of bone health and ID.

II. Development of advanced nurse practitioners and clinical nurse specialist in the area of bone health assessment and people with ID.

The development of advanced nurse practitioners/clinical nurse specialist (ANP/CNS) in the area of bone health is well established among the general population, however there is no such development within the ID field in Ireland. It is recognised and has been demonstrated that the addition of an ANP/CNS to the care package delivery and service provision is cost neutral and improves outcomes for clients/service users (Begley *et al.*, 2010). To that end the development of these roles would add value and benefit for service users, carer's and health care professionals alike in the following manner.

- To provide the necessary direct and indirect care and skills that will support those who care and support people with ID on an everyday basis.

- To liaise with the multidisciplinary team to support the bone health assessment of those with ID and advise on the appropriate course of action.

- As a conduit for the provision of audit, research and education to support all professionals in the area of bone health and people with ID.

- Bearing in mind that those with severe/profound level of ID may also be non-ambulant, have higher rates of co-morbidity and have greater dependence for care it is imperative that due consideration to level of ID is considered, an ANP/CNS would be in a position to advise, guide and support clinical practitioners on specifics regards ID and the impact on care.

- To provide inter and intra-disciplinary consultations, across all sites and services that effect or impact on the lives of people with ID.

III. Development of advocacy awareness groups for the promotion of health and well-being.

Irish health policy has shifted toward personal health responsibility, moving away from the perspective of illness toward the promotion of wellness. To that end the shift toward illness prevention and preservation of health requires promotion among people with ID. Being involved in the decision making process and thus gaining greater understanding of their own health is imperative, it is therefore essential that people with ID are involved in development, design and creation of health promotion. The researcher therefore proposes the development of a national interdisciplinary advocacy group, connecting the numerous supporting and networking groups for people with ID and the numerous advocacy groups throughout Ireland, in an endeavour to raise awareness of specific health challenges especially bone health. As part of the IDS-TILDA national study the researcher is positioned to promote this collaboration.

- Multidisciplinary approach to health care.
- Involvement of all stakeholders in the development health promotion campaigns.
- Health promotion campaigns that are accessible and easy read.

10.7.2 Future and further research recommendations

There are a number of areas in need of further investigation including strengthening the findings from this study through larger sampling, linking the data from this study with other similar studies being undertaken internationally, for example the Dutch HA study in the Erasmus Centre Rotterdam University, and, comparing this study with the TILDA Irish general population study.

Many areas for further research which merit investigation have been identified in this study and include; bone health among men with ID, bone health across the lifespan, biomedical and behavioural factors that contribute to fracture, fracture and its' impact on the lives of people with ID, lifestyle interventions that may prevent fracture, the need to identify and establish biomarker evidence for bone health, micro and macro nutrient impact on bone health, developing and validating physical activity and exercise programs for all people with ID especially those with difficulty walking and those with severe/profound ID as well as further research specifically targeting medication use and

the impact on bone health among adults with ID. The following agenda is provisionally proposed (see **table 10.1**). The list is preliminary and not exhaustive.

Table 10.1 Future potential research areas

Potential research area	Proposed research
Biological	What are the biomarkers of bone health and osteoporosis risk? Biomarkers and fracture incidence. Vitamin D deficiency among people with ID in Ireland. The researcher also intends to conduct a comparison study with the Irish general population study TILDA when their data becomes available.
Dietary health	What are the dietary behaviours of people with ID and how do these behaviours impact on bone health? What are the most effective micro and macro nutrient interventions to effect bone health? – consideration to vitamin supplementation Can improving dietary behaviour impact on bone health on a short or long term
Physical activity	Physical activity specific to bone health and level of ID? What is the influence of physical activity on the bone health of adults with ID? Can a physical activity intervention program improve bone health and/or prevent fracture?
Gender and bone health	Do men's (with ID) behaviours differ and so impact on their bone health? Are men with ID at higher risk of poor bone integrity and why? Osteoporosis among pre and postmenopausal female with ID.
Syndrome specific	Further examination of the factors that effect bone health among those with Down syndrome
Medication	An examination of the effects of AEDs on bone health among adults with ID An examination of the effects of PPIs on bone health of people with ID Medication cascade and its impact on bone health The impact of mental health drug use on bone health among people with ID
Economics	Assess the economic consequences of osteoporosis and fracture among people with ID

10.7.3 Future educational recommendations

The approach to the educational recommendations is divided into three sections. The perspective of the person with ID - adequate and appropriate education that is targeted for people with ID themselves; the perspective of their carer's and support workers who may or may not be trained health professionals and the perspective of the health professional with includes nurses, doctors, physiotherapist, dieticians, occupational therapist and any other health professional incorporated in the multidisciplinary team.

I. Specific for people with intellectual disability

- The involvement of people with ID in the development of positive health care packages for the improved care of their bone health.
- The development of appropriate educational packages in easy read format to support people with ID to understand the action required to maintain optimal bone health.
- Using of multimedia to deliver this message and promote positive health behaviours and actions. The researcher has traction in this area as she was involved in the development of the multimedia dissemination strategy for the IDS-TILDA study.

II. Specific for carer's and support workers of people with intellectual disability

- Development of educational packages to inform carer's of the positive requirements for excellent bone health among people with ID.
- Development of support packages to assist care's with supporting decision making among people with ID.
- Development of robust assessment guidelines to support carers in practice.

III. Specific for healthcare professionals

- Development of guidelines for healthcare professionals in relation to the specific bone health needs and requirements of people with ID.
- Particular attention to the educational needs around specific medication use in the field of ID and its impact on bone health.
- Development of assessment protocols and guidelines to support healthcare professionals in clinical practice.

10.7.4 Future policy recommendations

Raising political awareness of the needs and requirements of those more vulnerable, who are unable to do so themselves, is a civic responsibility. Lobbying is an essential part of this and to that end every endeavour ought to be made to increase the awareness of government representatives and agencies that are in a position to make policy change. To that end it is imperative as part of any research endeavour to have processes in place by which these public officials can be engaged to make the views and results of health research known, and in doing so better inform discussions and policy creation at local and national levels.

- Areas identified in this study in need of highlighting include the inclusion of the specific physical ability of people with ID and consideration of these abilities when operationalising health policy such as *Healthy Ireland (*HSE 2015*)*. The implementation of this policy is laid out in 3 clear priority strategies one of which is '*reducing the burden of chronic disease*' (HSE 2015: 15). And whilst this is a positive endeavour and in the whole applies to all citizens of Ireland the focus of chronic disease is very much on cancer and cardiovascular disease. However, the burden of disease patterns are different for people with ID subsequently they are marginalised in this policy's application. Therefore it is imperative to highlight areas such as the increased burden of poor bone health among people with ID.

- *Time to move on from congregated settings*: As the movement from congregated settings to the community occurs consideration is required for the unique health needs and requirements of people with ID, the educational requirements and support of their carer's and health professionals.

- The need to revisit the Food Safety Authority recommendations with regards vitamin D supplementation and raising awareness of these recommendations at national level.

- The need to address issues regards accessibility of health promotion. Whilst health promotion campaigns endeavour to be as accessible as possible this is not always the

case, it ought to be compulsory that all materials produced are targeted carefully for people with ID and developed with people with ID.

In summary the goal of Bone Health for Life would encompass the following four strategies in an endeavour to promote excellence in bone health specifically for people with ID, (see *figure 10.0).*

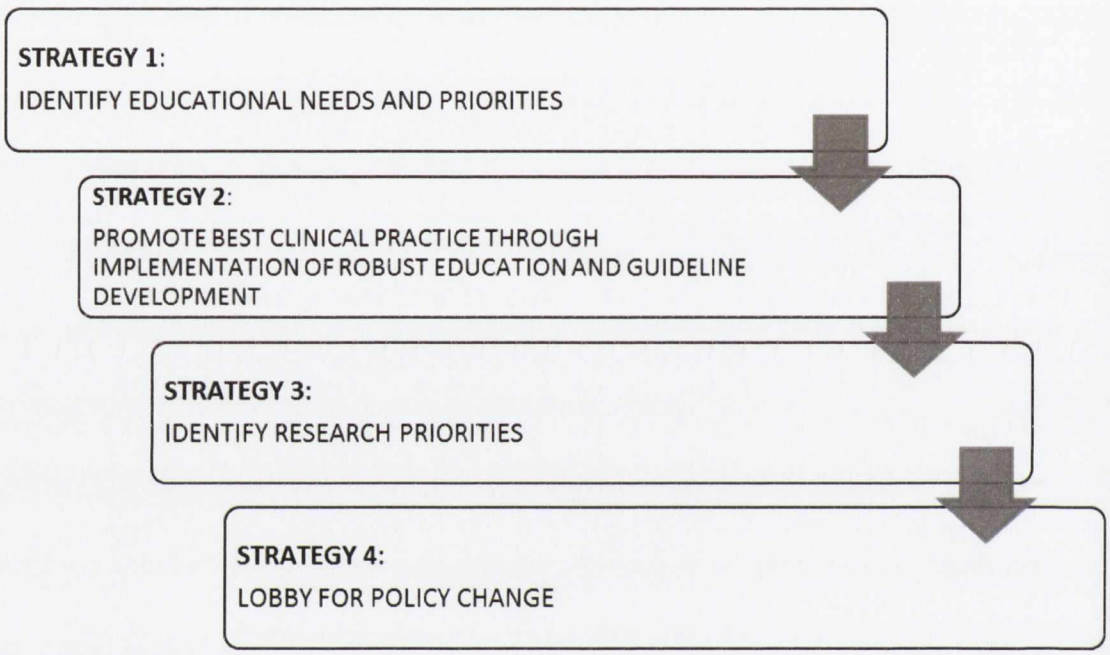
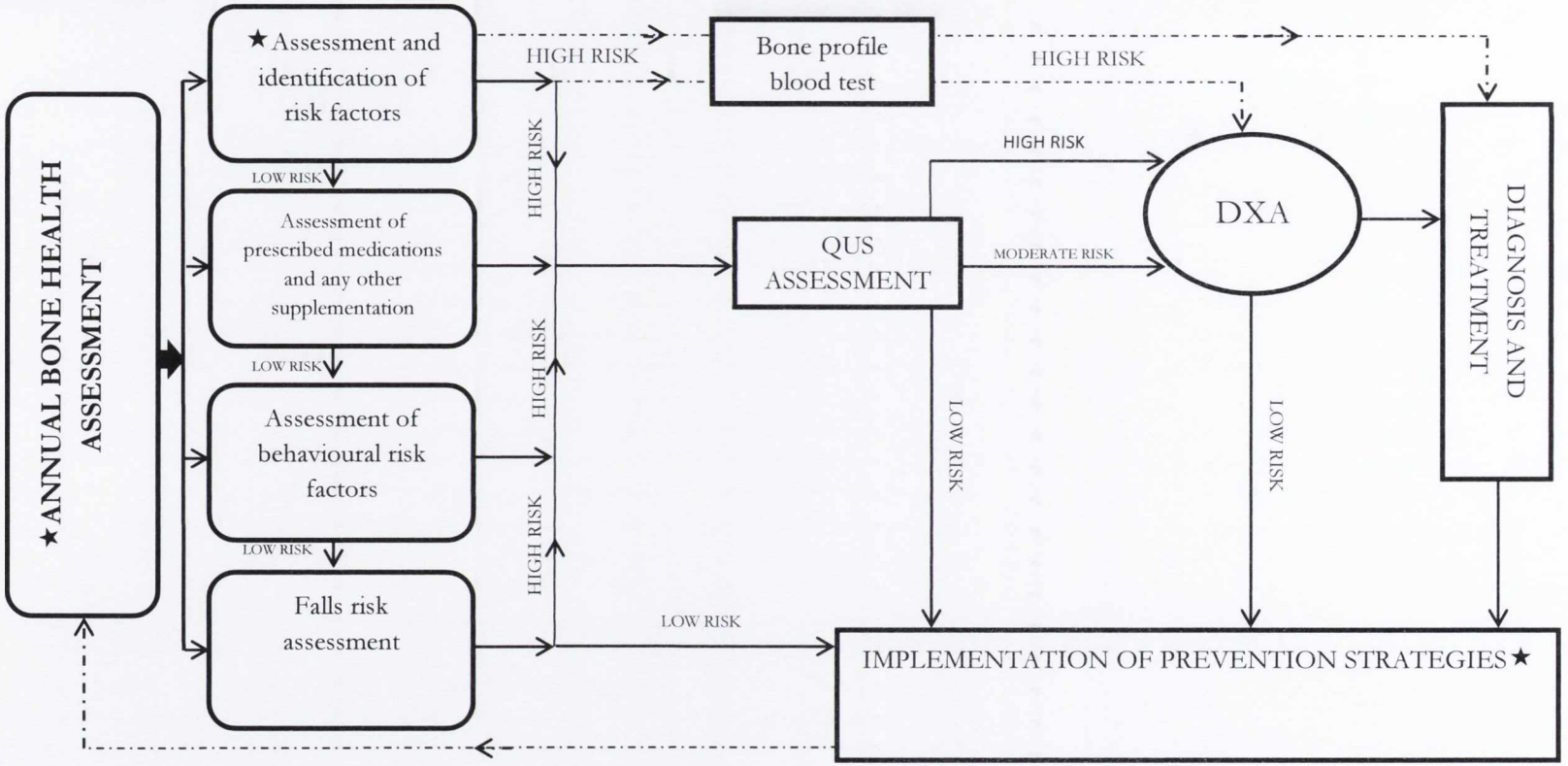


Figure 10.0 A strategy pathway for the promotion of bone health for life

10.7.5 Promoting the identification of osteopenia and osteoporosis

As part of the promotion of positive action to prevent and effectively treat osteopenia and osteoporosis among people with intellectual disability the researcher proposes the following armamentarium for clinical practice (see *figure 10.1*). This proposal is preliminary and will form the basis of further research among a larger sample of people with ID to develop a clinical decision pathway specifically for people with ID.

Depending on the clinical practitioner's decision the person with ID ought to have an annual robust bone health assessment. This assessment would incorporate examination of the commonly recognised risks, coupled with the specific risks associated with ID with particular attention to level of ID and the presence of co-morbidity, full medication review ought to be included with particular attention to the medications associated with bone integrity along with those in common use among people with ID (AEDs for example), an assessment of behavioural lifestyle would also be incorporated to include smoking, alcohol use, diet and vitamin supplementation and specific attention to physical activity levels and finally, a comprehensive falls risk assessment. The quantitative heel ultrasound could also contribute to the assessment as a quick non-invasive method of assessing bone quality and decisions on bone biochemistry could also be included for those identified at moderate to high risk. These would contribute to decisions on immediate treatment, further assessment in the form of DXA and the implementation of treatment and prevention strategies.



★Spotlights:

1 Assessment frequency would be determined by clinical practitioner, minimum recommendation annually, assessment would include all 4 domains

2 Further research is required to confirm specific risk factors among people with intellectual disability

3 Development of prevention strategies in the form of a multidisciplinary approach to include diet, behaviour lifestyle and physical activity

Figure 10.1 A bone health armamentarium for clinical practice

10.8 Conclusion

This study is the first comprehensive study on bone health among a large cohort of older adults with ID. The promotion of excellence in bone health is just one dimension of health care promotion that will contribute to improving the lives of people with ID. However, the insidious nature of osteoporosis makes it a particularly threatening condition that, without prevention and intervention, will have significant bearing on a person's life and quality of life. The biomechanical structural failure which results in osteoporosis is underpinned by a multitude of contributing factors. However, commonly identified risk factors observed among the general population such as smoking, alcohol, and corticosteroid use, are not consistently observed at rates to warrant concern among people with ID. This study maps out the increase and vulnerability of adults with ID for suboptimal bone health and identifies the unique risk factors among this population. Therefore, considering the unique findings in this study it is important when investigating the presentation of osteoporosis among people with ID to look beyond the 'common' contributing factors.

This study contributes to laying the foundations of building a greater understanding of the key determinants of bone health and well-being of adults with an ID as they grow older. However, to ensure a comprehensive approach to people's health consideration of the unique risk factors is just one component to realise optimal health. Cognitive skills to self-report are very often curtailed for people with ID impacting on the conversational interaction between them and their physician, understanding of health promotion may be restricted and very often the built environment lacks functionality and accessibility. Therefore, reasonable adjustment, of how the health message is transmitted, ensuring assessment and health promotion is delivered in an accessible manner for the individual and the support of the carer/support person of the person with ID are vital component for the smooth access and interpretation of peoples health needs to maximise their health now and to the future.

REFERENCES

Adolfsson, P., Sydner, Y. M., Fjellstrom, C., Lewin, B. & Andersson, A. (2008) Observed dietary intake in adults with intellectual disability living in the community. *Food Nutr Res,* **52**.

Åkesson, K., Vergnaud, P., Gineyts, E., Delmas, P. D. & Obrant, K. J. (1993) Impairment of bone turnover in elderly women with hip fracture. *Calcified tissue international,* **53**(3),162-169.

Alborz, A., McNally, R. & Glendinning, C. (2005) Access to health care for people with learning disabilities in the UK: mapping the issues and reviewing the evidence. *Journal of health services research & policy,* **10**(3), 173-182.

An Bord Altranais (2014) The code of professional conduct and ethics for each nurse and midwife. An Board Altranais, Dublin.

Anderson, F., Francis, R., Selby, P. & Cooper, C. (1998) Sex hormones and osteoporosis in men. *Calcified tissue international,* **62** (3), 185-188.

Angelopoulou, N., Matziari, C., Tsimaras, V., Sakadamis, A., Souftas, V. & Mandroukas, K. (2000) Bone mineral density and muscle strength in young men with mental retardation (with and without Down syndrome). *Calcif Tissue Int,* **66**(3), 176-80.

Angelopoulou, N., Souftas, V., Sakadamis, A. & Mandroukas, K. (1999) Bone mineral density in adults with Down's syndrome. *Eur Radiol,* **9**(4), 648-51.

Arvio, M., Kilpinen-Loisa, P., Tiitinen, A., Huovinen, K. & Makitie, O. (2009) Bone mineral density and sex hormone status in intellectually disabled women on progestin-induced amenorrhea. *Acta Obstetricia Et Gynecologica Scandinavica,* **88**(4), 428-433.

Aspray, T. J., Francis, R. M., Thompson, A., Quilliam, S. J., Rawlings, D. J. & Tyrer, S. P. (1998) Comparison of ultrasound measurements at the heel between adults with mental retardation and control subjects. *Bone,* **22**(9), 665-8.

Avenell, A., Gillespie, W., Gillespie, L. & O'Connell, D. (2009) Vitamin D and vitamin D analogues for preventing fractures associated with involutional and post-menopausal osteoporosis (Review). Wiley Online Library.

Avenell, A., Mak, J. & O'Connell, D. (2014) Vitamin D and vitamin D analogues for preventing fractures in post-menopausal women and older men. *Cochrane Database Syst Rev,* **4**.

Babayigit, A., Dirik, E., Bober, E. & Cakmakcı, H. (2006) Adverse effects of antiepileptic drugs on bone mineral density. *Pediatric neurology,* **35**(3), 177-181.

Babor, T., Higgins-Biddle, J., Saunders, J. & Monteiro, M. (2001) The Alcohol Use Disorders Identification Test, Guidelines for Use in Primary Care. Department of Mental Health and Substance Dependence, World Health Organization CH-1211 Geneva 27, Switzerland. ©World Health Organization

Balic, M. G., Mateos, E. C., Blasco, C. G. & Fernhall, B. (2000) Physical Fitness Levels of Physically Active and Sedentary Adults with Down Syndrome. *Adapted Physical Activity Quarterly,* **17**(3), 310-321.

Bannon, W. (2013) *The 7 Steps of Data Analysis: A Manual for Conduction a Quantitiative Research Study,* (Trans. StatsWhisperer Press, New York.

BAPEN (2012) Malnutrition Universal Screening Tool. Accessed: http://www.bapen.org.uk/. April 2013.

Baptista, F., Varela, A. & Sardinha, L. B. (2005) Bone mineral mass in males and females with and without Down syndrome. *Osteoporos Int,* **16**(4), 380-8.

Barnes, C. (1992) Qualitative Research: valuable or irrelevant, *Disability, Handiap and Society,* **7**(2), 115-124.

Barnes, C. (2002) 'Emancipatory disability research': project or process? *Journal of Research in Special Educational Needs,* **2**(1),

Barrett, A., Savva, G., Timonen, V. & Kenny, R. (2011) Fifty plus in Ireland 2011. *First results from the Irish Longitudinal Study on Ageing (TILDA). Trinity College Dublin, Dublin.*

Barrett-Connor, E., Siris, E. S., Wehren, L. E., Miller, P. D., Abbott, T. A., Berger, M. L., Santora, A. C. & Sherwood, L. M. (2005) Osteoporosis and fracture risk in women of different ethnic groups. *J Bone Miner Res,* **20**(2), 185-94.

Bastiaanse, L. P., Mergler, S., Evenhuis, H. M. & Echteld, M. A. (2014) Bone quality in older adults with intellectual disabilities. *Research in developmental disabilities,* **35**(9), 1927-1933.

Beange, H & Durvasula, S. (2001) Health inequalities in people with intellectual disability: Strategies for improvement. *Health Promotion Journal of Australia: Official Journal of Australian Association of Health Promotion Professionals,* **11**(1), 27.

Beange, H., Lennox, N. & Parmenter, T. R. (1999) Health targets for people with an intellectual disability. *Journal of Intellectual and Developmental Disability,* **24**(4), 283-297.

Beard, J. R., Biggs, S, Bloom, D E., Fried, L P., Hogan, P, Kalache, andOlshansky S. J (2011) Global Population Ageing: Peril or Promise,World Economic Forum. World Health Organisation, Geneva.

Beauchamp, T. L. & Childress, J. F. (2001) *Principles of biomedical ethics,* Oxford University press. London.

Beerhorst, K., de Krom, M., Aldenkamp, B., & Tan, F. (2009). Severe early onset osteoporosis in five young men with an intellectual disability, epilepsy and long-term antiepileptic drug use. *Epilepsia,* **50,** 77-77.

Begley C, Murphy K, Higgins A, Elliott N, Lalor J, Sheerin F, Coyne I, Comiskey C, Normand C, Casey C, Dowling M, Devane D, Cooney A, Farrelly F, Brennan M, Meskell P, MacNeela P. (2010) *Evaluation of Clinical Nurse and Midwife Specialist and Advanced Nurse and Midwife Practitioner Roles in Ireland (SCAPE) Final Report.* National Council for the Professional Development of Nursing and Midwifery in Ireland, Dublin.

Bergström, H., Elinder, L. & Wihlman, U. (2014) Barriers and facilitators in health education for adults with intellectual disabilities—a qualitative study. *Health education research,* **29**(2), 259-271.

Berkman, N. D., Sheridan, S. L., Donahue, K. E., Halpern, D. J. & Crotty, K. (2011) Low health literacy and health outcomes: an updated systematic review. *Annals of internal medicine,* **155**(2), 97-107.

Bittles, A. H, Bower, C., Hussain, R., & Glasson, E.J. (2007). The four ages of Down syndrome. *The European Journal of Public Health,* **17**(2), 221-225.

Bittles, A., Petterson, B., Sullivan, S., Hussain, R., Glasson, E. & Montgomery, P. (2002) The influence of intellectual disability on life expectancy. *The Journals of Gerontology Series A: Biological Sciences and Medical Sciences,* **57**(7), M470-M472.

Bliuc, D., Nguyen, N. D., Milch, V. E., Nguyen, T. V., Eisman, J. A. & Center, J. R. (2009) Mortality risk associated with low-trauma osteoporotic fracture and subsequent fracture in men and women. *JaMa,* **301**(5), 513-521.

Böhmer, C., Taminiau, J., Klinkenberg-Knol, E. & Meuwissen, S. (2001) The prevalence of constipation in institutionalized people with intellectual disability. *Journal of Intellectual Disability Research,* **45**(3), 212-218.

Bolton, J. M., Targownik, L. E., Leung, S., Sareen, J. & Leslie, W. D. (2011) Risk of low bone mineral density associated with psychotropic medications and mental disorders in postmenopausal women. *Journal of clinical psychopharmacology,* **31**(1), 56-60.

Braun, v. & Clarke V. (2006) Using thematic analysis in psychology. *Qualitative Research in Psychology,* **3,** 77-101.

Brenner. J (2008) FRAX: A Brand New Tool For the Management of Osteoporosis Clinical Correlations. . *Retrieved from [http://www.clinicalcorrelations.org/?p=818.] on 1 Feb 2012.*

Burge, R., Dawson-Hughes, B., Solomon, D. H., Wong, J. B., King, A. & Tosteson, A. (2007) Incidence and economic burden of osteoporosis-related fractures in the United States, 2005–2025. *Journal of bone and mineral research,* **22**(3), 465-475.

Burke, EA., McCallion, P. & McCarron, M. E. (2014) Advancing Years, Different Challenges: Wave 2 IDS-TILDA. Findings on the ageing of people with an intellectual disability. Trinity College Dublin, Ireland.

Burke, S. (2015) Boom to bust: its impact on Irish health policy and health services. Irish Journal of Public Policy Vol 2, 1 Accessed [http://publish.ucc.ie/ijpp/2010/01/burke/08/en] September 2015.

Burman, P., Ritzen, E. M. & Lindgren, A. C. (2001) Endocrine dysfunction in Prader-Willi syndrome: a review with special reference to GH. *Endocrine reviews,* **22**(6), 787-799.

Burns, N. & Grove, S. K. (2010) *Understanding nursing research: Building an evidence-based practice,* Elsevier Health Sciences.

Burt, D. B., Loveland, K. A., & Lewis, K. R. (1992). Depression and the onset of dementia in adults with mental retardation. *American Journal of Mental Retardation* **96**, 502-511.

Butts, J. B. & Rich, K. L. (2013) *Philosophies and theories for advanced nursing practice,* (Trans. Jones & Bartlett Publishers.

Carr, J. & Hollins, S. (1995) Menopause in women with learning disabilities. *Journal of Intellectual Disability Research,* **39**(2), 137-139.

Carroll, R., Burke, E., Mc Callion, P. & McCarron, M. (2014) Methodology. *IN: Advancing Years, Different Challenges: Findings on the Ageing of People with Intellectual Disability* (eds E. Burke, P. McCallion & M. McCarron). University of Dublin, Trinity College Dublin, Dublin.

Carter, N., Khan, K., Petit, M., Heinonen, A., Waterman, C., Donaldson, M., Janssen, P., Mallinson, A., Riddell, L. & Kruse, K. (2001) Results of a 10 week community based strength and balance training programme to reduce fall risk factors: a randomised controlled trial in 65–75 year old women with osteoporosis. *British Journal of Sports Medicine*, **35**(5) 348-351.

Castiglioni, S., Cazzaniga, A., Albisetti, W. & Maier, J. A. (2013) Magnesium and osteoporosis: current state of knowledge and future research directions. *Nutrients,* **5**(8), 3022-3033.

Center, J. R., McElduff, A. & Beange, H. (1994) Osteoporosis in groups with intellectual disability. *Australia & New Zealand Journal of Developmental Disabilities,* **19**(4), 251-258.

Center, J., Beange, H. & McElduff, A. (1998) People with mental retardation have an increased prevalence of osteoporosis: A population study. *American Journal on Mental Retardation,* **103**(1), 19-28.

Chew, K. L., Iacono, T. & Tracy, J. (2009) Overcoming communication barriers: Working with patients with intellectual disabilities. *Australian family physician,* **38**(1/2),10.

Collacott R.A., Cooper, S.-A. & McGrother, C. (1992) Differential rates of psychiatric disorders in adults with Down's syndrome compared with other mentally handicapped adults. *British Journal of Psychiatry,* **161**(13), 671–4.

Collinge, C. A., Lebus, G., Gardner, M. J. & Gehrig, L. (2010) A comparison of quantitative ultrasound of the calcaneus with dual-energy x-ray absorptiometry in hospitalized orthopaedic trauma patients. *Journal of orthopaedic trauma,* **24**(3), 176-180.

Compston, J. E., Watts, N. B., Chapurlat, R., Cooper, C., Boonen, S., Greenspan, S., Pfeilschifter, J., Silverman, S., Díez-Pérez, A. & Lindsay, R. (2011) Obesity is not protective against fracture in postmenopausal women: GLOW. *The American Journal of Medicine*, **124**(11), 1043-1050. Accessed at; http://www.iofbonehealth.org/guideline-references.

Compston, J., Bowring, C., Cooper, A., Cooper, C., Davies, C., Francis, R., Kanis, J., Marsh, D., McCloskey, E. & Reid, D. (2013) Diagnosis and management of osteoporosis in postmenopausal women and older men in the UK: National Osteoporosis Guideline Group (NOGG) update 2013. *Maturitas,* **75**(4), 392-396.

Compston, J., Cooper, A., Cooper, C., Francis, R., Kanis, J., Marsh, D., McCloskey, E., Reid, D., Selby, P. & Wilkins, M. (2009) Guidelines for the diagnosis and management of osteoporosis in postmenopausal women and men from the age of 50 years in the UK. *Maturitas,* **62**(2), 105-108.

Condon, D. (2002) Osteoporosis set to increase in Ireland Accessed at http://www.irishhealth.com/article.html?id=3931 March 2013.

Cooper, C. (2014) The Lifecourse Epidemiology of Musculoskeletal Ageing. In: *Osterporosis International.* pp. S101-S102. Springer London Ltd 236 Grays Inn rd., 6th floor, London WC1X 8HL, England.

Cooper, C., Cole, Z., Holroyd, C., Earl, S., Harvey, N., Dennison, E., Melton, L., Cummings, S. & Kanis, J. (2011) Secular trends in the incidence of hip and other osteoporotic fractures. *Osteoporos Int,* **22,** 1277 - 1288.

Cooper, S.-A., Melville, C. & Morrison, J. (2004) People with intellectual disabilities: their health needs differ and need to be recognised and met. *BMJ: British Medical Journal,* **329**(7463), 414.

Cooper, S.-A., Smiley, E., Morrison, J., Williamson, A. & Allan, L. (2007) Mental ill-health in adults with intellectual disabilities: prevalence and associated factors. *The British Journal of Psychiatry,* **190**(1), 27-35.

Corley, D. A., Kubo, A., Zhao, W. & Quesenberry, C. (2010) Proton pump inhibitors and histamine-2 receptor antagonists are associated with hip fractures among at-risk patients. *Gastroenterology,* **139**(1), 93-101.

Craig, C., Marshall, A., Sjöström, M., Bauman, A., Booth, M., Ainsworth, B., Pratt, M., Ekelund, U., Yngve, A. & Sallis, J. (2003) and the IPAQ Consensus Group and the IPAQ Reliability and Validity Study Group. International Physical Activity Questionnaire (IPAQ): 12-country reliability and validity. *Med Sci Sports Exerc,* **35,** 1381-1395.

Cronin, H., O'Regan, C. & Kenny, R. A. (2011) Physical and behavioural health of older Irish adults. *Fifty Plus in Ireland 2011,* 73.

Cullen, P. (2015) Broken; Irelands ailing health services. The Irish Times. Accessed at http://www.irishtimes.com/news/health/broken-ireland-s-ailing-health-service-1.2340468 September 2015.

da Silva, V. Z. M., de França Barros, J., de Azevedo, M., de Godoy, J. R. P., Arena, R. & Cipriano Jr, G. (2010) Bone mineral density and respiratory muscle strength in male individuals with mental retardation (with and without Down Syndrome). *Research in Developmental Disabilities,* **31**(6), 1585-1589.

Datta, H., Ng, W., Walker, J., Tuck, S. & Varanasi, S. (2008) The cell biology of bone metabolism. *Journal of clinical pathology,* **61**(5), 577-587.

de Asua, D. R., Parra, P., Costa, R., Moldenhauer, F., & Suarez, C. (2014). Evaluation of the impact of abdominal obesity on glucose and lipid metabolism disorders in adults with Down syndrome. *Research in Developmental Disabilities,* **35**(11), 2942-2949.

Department of Health (2013) Healthy Ireland, A framework for improved health and wellbeing 2013 - 2025. (ed Department of Health). Hawkins House, Dublin 2.

Department of Health and Children (DOHC) (2005) *Tackling Chronic Disease, A Policy Framework for the Management of Chronic Diseases,* Department of Health and Children, Hawkins House Dublin 2.

Dierssen, M., & De La Torre, R. (2012). *Down Syndrome: From Understanding the Neurobiology to Therapy* . Progress in Brain Research (Vol. 197): Elsevier Science, Oxford.

Doyle, M., Coughlan, T., Collins, R., & O'Neill, D. (2011). *Osteoporosis in Persons with Intellectual Disability: an initial screening programme.* Peamount Hospital, Adelaide and Meath Hospital, Tallaght, Ireland. CARDI.

Dreyfus, D., Lauer, E. & Wilkinson, J. (2014) Characteristics associated with bone mineral density screening in adults with intellectual disabilities. *The Journal of the American Board of Family Medicine,* **27**(1), 104-114.

Duffy, M. E. (2006) Handling missing data: a commonly encountered problem in quantitative research. *Clinical Nurse Specialist,* **20**(6), 273-276.

Ebeling, P. R. (2014) Osteoporosis in men, why change needs to happen. Accessed http://share.iofbonehealth.org/WOD/2014/thematic-report/WOD14-Report.pdf October 2014. (ed P. Mitchell). International Osteoporosis Foundation, Switzerland.

Elliott, J., Hatton, C. & Emerson, E. (2003) The health of people with learning disabilities in the UK: evidence and implications for the NHS. *Journal of Integrated Care,* **11**(3), 9-17.

Emerson, E. & Baines, S. (2011) Health inequalities and people with learning disabilities in the UK. *Tizard Learning Disability Review,* **16,** 42-48.

Emerson, E. & Hatton, C. (2013) *Health inequalities and people with intellectual disabilities,* (Trans. Cambridge University Press.

Emerson, E. (2005) Underweight, obesity and exercise among adults with intellectual disabilities in supported accommodation in Northern England. *Journal of Intellectual Disability Research,* **49**(Pt 2), 134-143.

Emerson, E., Robertson, J., Gregory, N., Kessissoglou, S., Hatton, C., Hallam, A., Knapp, M., Järbrink, K., Netten, A. & Linehan, C. (2000) The quality and costs of community-based residential supports and residential campuses for people with severe and complex disabilities. *Journal of Intellectual and Developmental Disability,* **25**(4), 263-279.

Evenhuis, H., Theunissen, M., Denkers, I., Verschuure, H. & Kemme, H. (2001) Prevalence of visual and hearing impairment in a Dutch institutionalized population with intellectual disability. *Journal of Intellectual Disability Research,* **45**(5), 457-464.

Felce, D., Baxter, H., Lowe, K., Dunstan, F., Houston, H., Jones, G., Grey, J., Felce, J. & Kerr, M. (2008) The impact of checking the health of adults with intellectual disabilities on primary care consultation rates, health promotion and contact with specialists. *Journal of Applied Research in Intellectual Disabilities,* **21**(6), 597-602.

Flaskerud, J. H. & Winslow, B. J. (1998) Conceptualizing Vulnerable Populations in Health Related Research *Nursing Research,* **47**(2), 69-78.

Food Safety Authority (2011) Scientific Recommendations for Healthy Eating Guidelines in Ireland. Food Safety Authority of Ireland, Dublin.

Foster, B. L., Walkley, J. W. & Temple, V. A. (2001) Bone mineral density status of women with intellectual disability. *Adapted Physical Activity Quarterly,* **18**(1), 49-59.

French, J. A. & Gazzola, D. M. (2011) New generation antiepileptic drugs: what do they offer in terms of improved tolerability and safety? *Therapeutic advances in drug safety*, 2042098611411127.

Frighi, V., Morovat, A., Stephenson, M.T., White, S.J., Hammond, C. V., & Goodwin, G.M. (2014). Vitamin D deficiency in patients with intellectual disabilities: prevalence, risk factors and management strategies. *The British Journal of Psychiatry,* **205**(6), 458-464.

Frost, J. (2015) The Danger of Overfitting Regression Models. In: *The Minitab Blog Accessed at http://blog.minitab.com/blog/adventures-in-statistics/the-danger-of-overfitting-regression-models Jan 2016.*

Fung, Y.-c. (2013) Bone and cartilage. In: *Biomechanics: mechanical properties of living tissues* pp. 389. Springer Science & Business Media, New York.

Gannon, B., O'Shea, E. & Hudson, E. (2007) The Economic Costs of falls and fractures in people aged 65 and over in Ireland. Irish Centre for Social Gerontology National University of Ireland, Galway

Gates, B. & Wilberforce, D. (2003) *The nature of learning disabilities (ed. Gates, Bob) Dimensions of Learning Disability,* Bailliere Tindall, London.

Gates, B. (2014) Learning disability nursing and people with profound intellectual disaiblities and complex needs. In: *Learning Disability Nursing: Modern Day Practice* (eds B. Gates & K. Mafuba). pp. 146. CRC Press, London.

Geijer, J. R., Stanish, H. I., Draheim, C. C., & Dengel, D. R. (2014). Bone mineral density in adults with Down syndrome, intellectual disability, and nondisabled adults. *American Journal on Intellectual & Developmental Disabilities,* **119**(2), 107-114. doi: 10.1352/1944-7558-119.2.107.

Genant, H. K., Cooper, C., Poor, G., Reid, I., Ehrlich, G., Kanis, J., Nordin, B. C., Barrett-Connor, E., Black, D. & Bonjour, J.-P. (1999) Interim report and recommendations of the World Health Organization task-force for osteoporosis. *Osteoporosis International,* **10**(4), 259-264.

Glasson, E.J, Dye, D.E, & Bittles, A.H. (2014). The triple challenges associated with age-related comorbidities in Down syndrome. *Journal of Intellectual Disability Research,* **58**(4), 393-398.

Glasson, E.J, Sullivan, S.G, Hussain, R, Petterson, B.A, Montgomery, P.D, & Bittles, A.H. (2002). The changing survival profile of people with Down's syndrome: implications for genetic counselling. *Clinical Genetics,* **62**(5), 390-393.

Glick, N. R., Fischer, M. H., Heisey, D. M., Leverson, G. E. & Mann, D. C. (2005) Epidemiology of fractures in people with severe and profound developmental disabilities. *Osteoporosis International,* **16**(4), 389-396.

Glüer, C. C., Eastell, R., Reid, D. M., Felsenberg, D., Roux, C., Barkmann, R., Timm, W., Blenk, T., Armbrecht, G. & Stewart, A. (2004) Association of five quantitative ultrasound devices and bone densitometry with osteoporotic vertebral fractures in a population-based sample: the OPUS Study. *Journal of Bone and Mineral Research,* **19**(5), 782-793.

Gold, E. B., Crawford, S. L., Avis, N. E., Crandall, C. J., Matthews, K. A., Waetjen, L. E., Lee, J. S., Thurston, R., Vuga, M. & Harlow, S. D. (2013) Factors related to age at natural menopause: longitudinal analyses from SWAN. *American journal of epidemiology*, kws421.

Gold, M., Franks, P. & Erickson, P. (1996) Assessing the health of the nation. The predictive validity of a preference-based measure and self-rated health. *Med Care,* **34**(2), 163-77.

Gonzalez-Aguero, A., Vicente-Rodriguez, G., Gomez-Cabello, A. & Casajus, J. A. (2013) Cortical and trabecular bone at the radius and tibia in male and female adolescents with Down syndrome: a peripheral quantitative computed tomography (pQCT) study. *Osteoporos Int,* **24**(3), 1035-44.

Gonzalez-Aguero, A., Vicente-Rodriguez, G., Moreno, L. A., & Casajus, J. A. (2011). Bone mass in male and female children and adolescents with Down syndrome. *Osteoporosis International,* **22**(7), 2151-2157. doi: 10.1007/s00198-010-1443-7

Government of Ireland (2003) Data Protection Act 1988 (Amendment), Amendment of Section 2 of the Principle Act. Available at: http://www.irishstatutebook.ie/2003/en/act/pub/0006/sec0003.html.

Grady, D. & Berkowitz, S. A. (2011) Why is a good clinical prediction rule so hard to find? *Archives of internal medicine,* **171**(19), 1701-1702.

Guessous, I., Cornuz, J., Ruffieux, C., Burckhardt, P. & Krieg, M. A. (2008) Osteoporotic fracture risk in elderly women: estimation with quantitative heel US and clinical risk factors. *Radiology,* **248**(1), 179-184.

Guijarro, M., Valero, C., Paule, B., Gonzalez-Macias, J., & Riancho, J. A. (2008). Bone mass in young adults with Down syndrome. *Journal of Intellectual Disability Research,* **52**(Pt 3), 182-189. doi: 10.1111/j.1365-2788.2007.00992.x

Hamann, C., Kirschner, S., Günther, K.-P. & Hofbauer, L. C. (2012) Bone, sweet bone—osteoporotic fractures in diabetes mellitus. *Nature Reviews Endocrinology,* **8,** 297-305.

Hans, D., Dargent-Molina, P., Schott, A., Sebert, J., Cormier, C., Kotzki, P., Delmas, P., Pouilles, J., Breart, G. & Meunier, P. (1996) Ultrasonographic heel measurements to predict hip fractures in elderly women: the EPIDOS prospective study. *Lancet,* **348**(9026) 511 - 514.

Hans, D., Hartl, F. & Krieg, M. (2003) Device-specific weighted T-score for two quantitative ultrasounds: operational propositions for the management of osteoporosis for 65 years and older women in Switzerland. *Osteoporosis international,* **14**(3), 251-258.

Hatton, C., Roberts, H. & Baines, S. (2011) Reasonable adjustments for people with learning disabilities in England 2010: A national survey of NHS Trusts. *Durham: Improving Health & Lives: Learning Disabilities Observatory.*

Haveman, M., Heller, T., Lee, L., Maaskant, M., Shooshtari, S., & Strydom, A. (2010). Major health risks in aging persons with intellectual disabilities: an overview of recent studies. *Journal of Policy and Practice in Intellectual Disabilities,* **7**(1), 59-69.

Haveman, M., Perry, J., Salvador-Carulla, L., Walsh, P. N., Kerr, M., Van Schrojenstein Lantman-de Valk, H., Van Hove, G., Berger, D. M., Azema, B., Buono, S., Cara, A. C., Germanavicius, A., Linehan, C., Määttä, T., Tossebro, J. & Weber, G. (2011) Ageing and health status in adults with intellectual disabilities: Results of the European POMONA II study. *Journal of Intellectual and Developmental Disability,* **36**(1), 49-60.

Hayes, A. & Batshaw, M. L. (1993) Down syndrome. *Pediatric Clinics of North America,* **40**(3), 523-535.

Health Service Executive (2008) Strategy to prevent falls and fractures in Ireland, summary, conclusions and recommendations. HSE, Dublin.

Health Service Executive (2010) Get Ireland Active, Promoting Physical Activity in Ireland. HSE, Dublin.

Health Service Executive (2011) Time to Move on from Congregated Settings A Strategy for Community Inclusion. In: *Report of the Working Group on Congregated Settings.* HSE, Dublin.

Health Service Executive (2012) Healthy Eating and Active Living for Adults,Teenagers and Children over 5 Years – A Food Guide for Health Professionals and Catering Services. Food Safety Authority of Ireland Abbey Court, Lower Abbey Street, Dublin 1.

Health Service Executive (2015) Healthy Ireland in the Health Services National Implementation Plan 2015 – 2017. Retrieved from : http://www.hse.ie/eng/health/hl/hi/HIDocs/HImplementationplanWeb.pdf. Health Service Executive, Dublin.

Health Service Executive (HSE) (2008) Strategy to Prevent Falls and Fractures in Ireland's Ageing Population. Report of the National Steering Group on the Prevention of Falls in Older People and the Prevention and Management of Osteoporosis throughout Life. In: *Strategic Health Planning.* Dr Steeven's Hospital, Dublin 8.

Health Service Executive (HSE), & Department of Health and Children (DOHC). (2012). Your guide to healthy eating using the food pyramid. Retrieved from http://www.lenus.ie/hse/handle/10147/305177 January 2014.

Health Research Board (2013). Retrieved from http://www.hrb.ie/health-information-in-house-research/disability/nidd/April 2013.

Heller, T., Hsieh, K., & Rimmer, J. (2003). Barriers and supports for exercise participation among adults with Down syndrome. *Journal of Gerontological Social Work,* **38**(1-2), 161-178.

Henderson, R. C., Lark, R. K., Gurka, M. J., Worley, G., Fung, E. B., Conaway, M., Stallings, V. A. & Stevenson, R. D. (2002) Bone density and metabolism in children and adolescents with moderate to severe cerebral palsy. *Pediatrics,* **110**(1), e5-e5.

Hernlund, E., Svedbom, A., Ivergård, M., Compston, J., Cooper, C., Stenmark, J., McCloskey, E., Jönsson, B. & Kanis, J. (2013) Osteoporosis in the European Union: medical management, epidemiology and economic burden. *Archives of osteoporosis,* **8**(1-2), 1-115. http://www.irishosteoporosis.ie/images/uploads/Osteoporosis-Guidelines.pdf, Dublin, Ireland. Accessed March 2013.

Herrera, A., Lobo-Escolar, A., Mateo, J., Gil, J., Ibarz, E. & Gracia, L. (2012) Male osteoporosis: A review. *World journal of orthopedics,* **3**(12), 223.

Heslop, P., Blair, P. S., Fleming, P., Hoghton, M., Marriott, A. & Russ, L. (2014) The Confidential Inquiry into premature deaths of people with intellectual disabilities in the UK: a population-based study. *The Lancet,* **383**(9920), 889-895.

Heslop, P., Blair, P., Fleming, P., Hoghton, M., Marriott, A. & Russ, L. (2007) Confidential Inquiry into premature deaths of people with learning disabilities (CIPOLD). Bristol, England: Norah Fry Research Centre. resources for persons with intellectual disabilities.

HIPE (2013) Health Research and Information Division, Economic and Social Research Institute, . *Hospital In-Patient Enquiry Scheme, Data Dictionary 2013 Version 5.0 Accessed at: http://www.hpo.ie/hipe/hipe_data_dictionary/HIPE_Data_Dictionary_2013_V5.0.pdf,* Oct 2015

Hirani, V. & Primatesta, P. (2005) Vitamin D concentrations among people aged 65 years and over living in private households and institutions in England: population survey. *Age and Ageing,* **34**(5), 485-491.

Hofbauer, L. C., Brueck, C. C., Singh, S. K. & Dobnig, H. (2007) Osteoporosis in patients with diabetes mellitus. *Journal of Bone and Mineral Research,* **22**(9), 1317-1328.

Holick, M. F. (2003) Vitamin D: A millenium perspective. *J Cell Biochem,* **88**(2), 296-307.

Holloway, K., Brennan, S., Kotowicz, M., Bucki-Smith, G., Timney, E., Dobbins, A., Williams, L. & Pasco, J. (2015) Prior fracture as a risk factor for future fracture in an Australian cohort. *Osteoporosis International,* **26**(2), 629-635.

Hsiang, Y. H. H., Berkovitz, G. D., Bland, G. L., Migeon, C. J., Warren, A. C., Opitz, J. M. & Reynolds, J. F. (1987) Gonadal function in patients with Down syndrome. *American journal of medical genetics,* **27**(2), 449-458. IBM Corp. Released 2013.Accessed at: http://www.irishosteoporosis.ie/images/uploads/Osteoporosis-Guidelines.pdf, Dublin, Ireland. March 2013.

Hunt, C., Wakefield, S. & Hunt, G. (2001) Community nurse learning disabilities: a case study of the use of an evidence-based screening tool to identify and meet the health needs of people with learning disabilities. *Journal of Intellectual Disabilities,* **5**(1), 9-18.

Iacono, T. & Sutherland, G. (2006) Health screening and developmental disabilities. *Journal of Policy and Practice in Intellectual Disabilities,* **3,** 155-163.

IBM (2013) IBM Corp. Released 2013. IBM SPSS Data Collection. Armonk, NY: IBM Corporation.

IBM (2011) IBM SPSS Decision 20. Accessed at https://www.csun.edu/sites/default/files/decision-trees20-64bit.pdf January 2016. (ed SPSS Inc. 1989). IBM Corporation, , North Castle Drive, Armonk, NY 10504-1785.

Idhammad, A. & Abdali, A. (2012) On a new law of bone remodeling based on damage elasticity: a thermodynamic approach. *Theor Biol Med Model,* **9,** 51.

Iezzoni, L. I., Kilbridge, K. & Park, E. (2010) Physical Access Barriers to Care for Diagnosis and Treatment of Breast Cancer Among Women with Mobility Impairments. *Oncol Nurs Forum,* **37**(6), 711-717.

Iezzoni, L. I., McCarthy, E. P., Davis, R. B. & Siebens, H. (2000) Mobility impairments and use of screening and preventive services. *American Journal of Public Health,* **90**(6), 955.

International Osteoporosis Foundation (IOF) (2012) Capture the Fracture: a global campaign to break the fragility fracture cycle. http://www.worldosteoporosisday.org/ Accessed 01.07.2015

Irish Osteoporosis Society (2012) Osteoporosis Guidelines for Health Professionals. Irish Osteoporosis Society Accesssed at; https://www.icgp-education.ie/osteoporosis/Osteoporosis-Soc-Guidelines-2011.pdf, March 2013

Ito, T. & Jensen, R. T. (2010) Association of long-term proton pump inhibitor therapy with bone fractures and effects on absorption of calcium, vitamin B12, iron, and magnesium. *Current gastroenterology reports,* **12**(6), 448-457.

Jaffe, J. S., Timell, A. M. & Gulanski, B. I. (2001) Prevalence of low bone density in women with developmental disabilities. *J Clin Densitom,* **4**(1), 25-9.

Jaffe, J. S., Timell, A. M., Elolia, R. & Thatcher, S. S. (2005) Risk factors for low bone mineral density in individuals residing in a facility for the people with intellectual disability. *Journal of Intellectual Disability Research,* **49**(6), 457-462.

Janicki, M. P., Dalton, A. J., Henderson, C. M. & Davidson, P. W. (1999) Mortality and morbidity among older adults with intellectual disability: health services considerations. *Disability & Rehabilitation,* **21**(5-6), 284-294.

Jaovisidha, S., Kim, J. K., Sartoris, D. J., Bosch, E., Edelstein, S., Barrett-Connor, E. & Rojanaplakorn, P. (1998) Scoliosis in elderly and age-related bone loss: a population-based study. *Journal of Clinical Densitometry,* **1**(3), 227-233.

Jasien, J., Daimon, C. M., Maudsley, S., Shapiro, B. K. & Martin, B. (2012) Aging and bone health in individuals with developmental disabilities. *Int J Endocrinol,* **2012,** 469235.

Johnell, O. & Kanis, J. (2006) An estimate of the worldwide prevalence and disability associated with osteoporotic fractures. *Osteoporosis international,* **17**(12),1726-1733.

Kanis, J. (1994) Assessment of fracture risk and its application to screening for postmenopausal osteoporosis: synopsis of a WHO report. *Osteoporosis International,* **4**(6), 368-381.

Kanis, J. A. (2002) Diagnosis of osteoporosis and assessment of fracture risk. *The Lancet,* **359,** (9321)1929-1936.

Kanis, J. A., Johansson, H., Oden, A. & McCloskey, E. V. (2009a) Assessment of fracture risk. *European Journal of Radiology,* **71,** 392-397.

Kanis, J. A., Oden, A., Johansson, H., Borgström, F., Ström, O. & McCloskey, E. (2009b) FRAX< sup>®</sup> and its applications to clinical practice. *Bone,* **44,** 734-743

Kanis, J. A., Odén, A., McCloskey, E., Johansson, H., Wahl, D. A. & Cooper, C. (2012) A systematic review of hip fracture incidence and probability of fracture worldwide. *Osteoporosis International,* **23,** 2239-2256.

Kanis, J., Black, D., Cooper, C., Dargent, P., Dawson-Hughes, B., De Laet, C., Delmas, P., Eisman, J., Johnell, O. & Jonsson, B. (2002) A new approach to the development of assessment guidelines for osteoporosis. *Osteoporosis International,* **13**(7), 527-536.

Kanis, J., Burlet, N., Cooper, C., Delmas, P., Reginster, J.-Y., Borgstrom, F. & Rizzoli, R. (2008a) European guidance for the diagnosis and management of osteoporosis in postmenopausal women. *Osteoporosis international,* **19**(4), 399-428.

Kanis, J., Johnell, O., Odén, A., Johansson, H. & McCloskey, E. (2008b) FRAX™ and the assessment of fracture probability in men and women from the UK. *Osteoporosis International,* **19**(4), 385-397.

Kanis, J., McCloskey, E., Johansson, H., Cooper, C., Rizzoli, R. & Reginster, J.-Y. (2013) European guidance for the diagnosis and management of osteoporosis in postmenopausal women. *Osteoporosis International,* **24**(1), 23-57.

Kanis, J., Odén, A., Johnell, O., Johansson, H., De Laet, C., Brown, J., Burckhardt, P., Cooper, C., Christiansen, C. & Cummings, S. (2007) The use of clinical risk factors enhances the performance of BMD in the prediction of hip and osteoporotic fractures in men and women. *Osteoporosis international,* **18**(8),1033-1046.

Kanis, J.A, Melton, L .J., Christiansen, C., Johnston, C.C., & Khaltaev, N. (1994). The diagnosis of osteoporosis. *Journal of Bone and Mineral Research,* **9**(8), 1137-1141

Kao, C.H., Chen, C.C., Wang, S.J., & Yeh, S.H. (1992). Bone mineral density in children with Down's syndrome detected by dual photon absorptiometry. *Nuclear Medicine communications,* **13**(10), 773-775.

Karinkanta, S., Heinonen, A., Sievänen, H., Uusi-Rasi, K., Pasanen, M., Ojala, K., Fogelholm, M. & Kannus, P. (2007) A multi-component exercise regimen to prevent functional decline and bone fragility in home-dwelling elderly women: randomized, controlled trial. *Osteoporosis International,* **18**(4), 453-462.

Karlsson, B., Gustafsson, J., Hedov, G., Ivarsson., S.A., & Annerén, G. (1998). Thyroid dysfunction in Down's syndrome: relation to age and thyroid autoimmunity. *Archives of Disease in Childhood,* **79**(3), 242-245.

Kass, G. V. (1980) An exploratory technique for investigating large quantities of categorical data. *Applied statistics,* 119-127.

Katterl, R. & Bywood, P. T. (2011) Primary health care for people with intellectual disabilities. *Research Roundup; Primary Health Care Reserach and Information Service,* **17**.

Kavanagh, R. (2015) Osteoporosis; A disease on the rise. Accessed at http://www.irishtimes.com/life-and-style/the-health-centre/osteoporosis/osteoporosis-a-disease-on-the-rise-1.2058970. Report for the Irish Times Jan 14, 2015.

Kelly F., Kelly C. & Craig S. (2009) HRB Statistics Series: Annual Report of the National Intellectual Disability Database Committee. Health Research Board, Dublin.

Kelly, C. & O'Donohoe, M. (2014) *Annual report of the National Intellectual Disability Database Committee 2013.* Health Research Board, Dublin, Ireland.

Kenny, R. & Nolan, A. (2014) *The Irish Longitudinal Study on Ageing Wave 2: The Over 50's in a Changing Ireland, Economic Circumstances Health and Wellbeing.* Trinity College Dublin, Dublin.

Kenny, R. A., Whelan, B. J., Cronin, H., Kamiya, Y., Kearney, P., O'Regan, C. & Ziege, M. (2010) The Design of the Irish Longitudinal Study on Ageing. (eds A. Barrett, C. Finucane & V. Timonen). Universtiy of Dublin, Trinity College Dublin, Dublin.

Kerr, M. (1998) Primary health care and health gain for people with a learning disability. *Tizard Learning Disability Review,* **3**(4), 6-14.

Kerr, M. (2004) Improving the general health of people with learning disabilities. *Advances in Psychiatric Treatment,* **10**(3), 200-206.

Kevan, F. (2003) Challenging behaviour and communication difficulties. *British Journal of Learning Disabilities,* **31,** 75-80.

Khaw, K.-T., Reeve, J., Luben, R., Bingham, S., Welch, A., Wareham, N., Oakes, S. & Day, N. (2004) Prediction of total and hip fracture risk in men and women by quantitative ultrasound of the calcaneus: EPIC-Norfolk prospective population study. *The Lancet,* **363**(9404), 197-202.

Kirschner, K. L., Breslin, M. L. & Iezzoni, L. I. (2007) Structural impairments that limit access to health care for patients with disabilities. *Journal of the American Medical Association.* **297**(10), 1121-1125.

Kluger, A. (2011) Inclusive design. Architecture for everyone.*Accessed at*: https://www.politesi.polimi.it/bitstream/10589/19143/1/Inclusive%20design%20-%20Architecture%20for%20Everyone.pdf. July 2014.

Knickmeyer, R. C., Wheelwright, S., Hoekstra, R. & Baron-Cohen, S. (2006) Age of menarche in females with autism spectrum conditions. *Developmental Medicine & Child Neurology,* **48**(12), 1007-1008.

Kohrt, W. M., Ehsani, A. A. & Birge, S. J. (1997) Effects of Exercise Involving Predominantly Either Joint-Reaction or Ground-Reaction Forces on Bone Mineral Density in Older Women. *Journal of Bone and Mineral Research,* **12**(8), 1253-1261.

Krahn, G. L., Hammond, L. & Turner, A. (2006) A cascade of disparities: health and health care access for people with intellectual disabilities. *Mental retardation and developmental disabilities research reviews,* **12**(1), 70-82.

Krassas, G.E., & Papadopoulou, P.H. (2001). Oestrogen action on bone cells. *Journal of Musculoskeletal and Neuronal Interactions,* **2**(2), 143-152.

Krieg, M.-A., Barkmann, R., Gonnelli, S., Stewart, A., Bauer, D. C., Del Rio Barquero, L., Kaufman, J. J., Lorenc, R., Miller, P. D. & Olszynski, W. P. (2008) Quantitative ultrasound in the management of osteoporosis: the 2007 ISCD Official Positions. *Journal of Clinical Densitometry,* **11,** 163-187.

Lane, J. M., Serota, A. C. & Raphael, B. (2006) Osteoporosis: differences and similarities in male and female patients. *Orthopedic Clinics of North America,* **37**(4), 601-609.

Lee, C. H., Lee, M. K., Han, H. J., Kim, T. H., Lee, J. H. & Kim, S. H. (2013) Response: The Association between Serum Endogenous Secretory Receptor for Advanced Glycation End Products and Vertebral Fractures in Type 2 Diabetes (Endocrinol Metab 2012; 27: 289-94, Cheol Ho Lee *et al.*,). *Endocrinology and Metabolism,* **28**(1), 78-79.

Lennox, N. & Kerr, M. (1997) Primary health care and people with an intellectual disability: the evidence base. *Journal of Intellectual Disability Research,* **41**(5), 365-372.

Lennox, N., Diggens, J. & Ugoni, A. (1997) The general practice care of people with intellectual disability: barriers and solutions. *Journal of Intellectual Disability Research,* **41**(5), 380-390.

Leslie, W. D., Pahlavan, P. S., Roe, E. B. & Dittberner, K. (2009) Bone density and fragility fractures in patients with developmental disabilities. *Osteoporosis International,* **20**(3), 379-383.

Lewiecki, E. M., Richmond, B. & Miller, P. D. (2006) Uses and misuses of quantitative ultrasonography in managing osteoporosis. *Cleveland Clinic journal of medicine,* **73**(8), 742.

Lewis, C., Robertson, A. S. & Phelps, S. (2005) Unmet dental care needs among children with special health care needs: implications for the medical home. *Pediatrics,* **116**(3), e426-e431.

Lin, L.-P., Hsu, S.-W., Yao, C.-H., Lai, W.-J., Hsu, P.-J., Wu, J.-L., Chu, C. M. & Lin, J.-D. (2015) Risk for osteopenia and osteoporosis in institution-dwelling individuals with intellectual and/or developmental disabilities. *Research in Developmental Disabilities,* **36,** 108-113.

Linehan, C., Kerr, M. P., Walsh, P. N., Brady, G., Kelleher, C., Delanty, N., Dawson, F. & Glynn, M. (2010) Examining the prevalence of epilepsy and delivery of epilepsy care in Ireland. *Epilepsia,* **51**(5), 845-852.

Lohiya, G.-S., Tan-Figueroa, L. & Iannucci, A. (2004) Identification of low bone mass in a developmental center: finger bone mineral density measurement in 562 residents. *Journal of the American Medical Directors Association,* **5**(6), 371-376.

Lord, S. R., Smith, S. T., & Menant, J. C. (2010). Vision and falls in older people: Risk factors and intervention strategies. Clinics in Geriatric Medicine, **2**(6), 569-581.

Mac Giolla Phadraig, C., McCallion, P., Cleary, E., McGlinchey, E., Burke, E., McCarron, M. & Nunn, J. (2014) Total tooth loss and complete denture use in older adults with intellectual disabilities in Ireland. *Journal of public health dentistry.* Access at: http://onlinelibrary.wiley.com.elib.tcd.ie/doi/10.1111/jphd.12077/epdf

Martin, D. M., Cassidy, G., Ahmad, F. & Martin, M. S. (2001) Women with learning diabilities and the menopause. *Journal of Learning Disabilities,* **5**(2), 121-132.

Martin, G. (2003) Annual Health Reviews for Patients with Severe Learning Disabilities: Five Years of a Combined GP/CLDN Clinic. *Journal of Intellectual Disabilities,* **7**(1), 9-21.

Martínez-Leal, R., Salvador-Carulla, L., Linehan, C., Walsh, P., Weber, G., Van Hove, G., Määttä, T., Azema, B., Haveman, M., Buono, S., Germanavicius, A., van Schrojenstein Lantman-de Valk, H., Tossebro, J., Carmen-Câra, A., Moravec Berger, D., Perry, J. & Kerr, M. (2011) The impact of living arrangements and deinstitutionalisation in the health status of persons with intellectual disability in Europe. *Journal of Intellectual Disability Research,* **55**(9), 858-872.

Mathiowetz, V., Kashman, N., Volland, G., Weber, K., Dowe, M. & Rogers, S. (1985) Grip and pinch strength: normative data for adults. *Archives of physical medicine and rehabilitation,* **66**(2), 69-74.

Matignon, R. (2007) *Data mining using SAS enterprise miner,* John Wiley & Sons.

Matsuzaki, H., Katsumata, S.-i., Kajita, Y. & Miwa, M. (2013) Magnesium deficiency regulates vitamin D metabolizing enzymes and type II sodium-phosphate cotransporter mRNA expression in rats. *Magnesium Research,* **26**(2), 83-86.

Mattson, R. H. & Gidal, B. E. (2004) Fractures, epilepsy, and antiepileptic drugs. *Epilepsy & Behavior,* **5,** 36-40.

Matute-Llorente, A., Gonzalez-Aguero, A., Gomez-Cabello, A., Vicente-Rodriguez, G., & Casajus, J. A. (2013). Decreased levels of physical activity in adolescents with Down

syndrome are related with low bone mineral density: a cross-sectional study. *BMC Endocrine Disorders,* **13**(1), 22. doi: 10.1186/1472-6823-13-22.

McCallion P. & McCarron M. (2004) Aging and Intellectual disabilities: a review of recent literature. *Current Opinion in Psychiatry* **17**, 349–52.

McCarron, M., & Lawlor, B.A. (2003). Responding to the challenges of ageing and dementia in intellectual disability in Ireland. *Aging & Mental Health,* **7**(6), 413-417.

McCarron, M., Burke, E., Cleary, E., Carroll, R., McGlinchey, E. & McCallion, P. (2014) Changes in Physical and Behavioural Health of Older Adults with Intellectual Disability. IN: *Advancing Years Different Challenges* (eds E. Burke, P. McCallion & M. McCarron). University of Dublin, Trinity College Dublin.

McCarron, M., Carroll, R., Kelly, C. & McCallion, P. (2015) Mortality Rates in the General Irish Population Compared to those with an Intellectual Disability from 2003 to 2012. *Journal of Applied Research in Intellectual Disabilities,* **28**(5), 406-413.

McCarron, M., Gill, M., McCallion, P., & Begley, C. (2005). Health co-morbidities in ageing persons with Down syndrome and Alzheimer's dementia. *Journal of Intellectual Disability Research,* **49**(7), 560-566.

McCarron, M., McCallion, P., Reilly, E. & Mulryan, N. (2013b) A prospective 14-year longitudinal follow-up of dementia in persons with Down syndrome. *Journal of Intellectual Disability Research.* **58**(1), 61-70.

McCarron, M., Swinburne, J., Andrews, V., McGarvey, B., Murray, M., Mulryan, N., Drummond, E., Sherrin, F. & McCallion, P. (2009) Intellectual Disability Supplement to The Irish Longitudinal Study of Ageing. Pilot Report. School of Nursing & Midwifery, Trinity College Dublin.

McCarron, M., Swinburne, J., Burke, E., McGlinchey, E., Carroll, R. & McCallion, P. (2013a) Patterns of multimorbidity in an older population of persons with an intellectual disability: results from the intellectual disability supplement to the Irish longitudinal study on aging (IDS-TILDA). *Research in developmental disabilities,* **34**(1), 521-527.

McCarron, M., Swinburne, J., Burke, E., McGlinchey, E., Mulryan, N., Andrews, V., Foran, S. & McCallion, P. (2011). *Growing Older with an Intellectual Disability in Ireland 2011: First Results from the Intellectual Disability Supplement to the Irish Longitudinal Study on Ageing (IDS-TILDA)*: School of Nursing and Midwifery, Trinity College Dublin.

McCausland, D., McCallion, P., Cleary, E. & McCarron, M. (2015) Social Connections for Older People with Intellectual Disability in Ireland: Results from Wave One of IDS-TILDA. *Journal of Applied Research in Intellectual Disabilities.* **29,** 71-82.

McKelvey, K. D., Fowler, T. W., Akel, N. S., Kelsay, J. A., Gaddy, D., Wenger, G. R., & Suva, L. J. (2013a). Low bone turnover and low bone density in a cohort of adults with Down syndrome. *Osteoporosis International,* **24**(4), 1333-1338. doi: 10.1007/s00198-012-2109-4

Melton, L. J. (2003) Adverse outcomes of osteoporotic fractures in the general population. *Journal of Bone and Mineral Research,* **18**(6), 1139-1141.

Melville, C. A., Hamilton, S., Hankey, C. R., Miller, S. & Boyle, S. (2007) The prevalence and determinants of obesity in adults with intellectual disabilities. *Obes Rev,* **8**(3), 223-30.

Melville, C., Cooper, S. A., Morrison, J., Finlayson, J., Allan, L., Robinson, N., Burns, E. & Martin, G. (2006) The outcomes of an intervention study to reduce the barriers experienced by people with intellectual disabilities accessing primary health care services. *Journal of intellectual disability research,* **50**(1), 11-17.

Mergler, S. (2012) *Bone Quality in Children with Severe Neurological Impairment and Intellectual Disability: Diagnostic methods and determinants of low bone quality,* Erasmus MC: University Medical Center Rotterdam.

Mergler, S., Bastiaanse, L., Echteld, M. & Evenhuis, H. (2012) Prevalence and associated factors of osteoporosis in older adults with intellectual disabilties. In: *Journal of Intellectual Disability Research.* pp. 757-757. Wiley-Blackwell 111 River St, Hoboken 07030-5774, NJ USA.

Mergler, S., Evenhuis, H. M., Boot, A. M., De Man, S. A., Bindels-De Heus, K. G. C. B., Huijbers, W. A. R. & Penning, C. (2009) Epidemiology of low bone mineral density and fractures in children with severe cerebral palsy: A systematic review. *Developmental Medicine & Child Neurology,* **51**(10), 773-778.

Mergler, S., Lobker, B., Evenhuis, H. M. & Penning, C. (2010) Feasibility of quantitative ultrasound measurement of the heel bone in people with intellectual disabilities. *Research in Developmental Disabilities,* **31**(6), 1283-1290.

Meunier, P. J., Delmas, P. D., Eastell, R., McClung, M. R., Papapoulos, S., Rizzoli, R., Seeman, E. & Wasnich, R. D. (1999) Diagnosis and management of osteoporosis in postmenopausal women: clinical guidelines. *Clinical Therapeutics,* **21**(6), 1025-1044.

Michael, J. (2008) Healthcare for all. *Report of the independent inquiry into access to healthcare for people with learning disabilities. London: Department of Health.*

Mikati, M., Dib, L., Yamout, B., Sawaya, R., Rahi, A. & Fuleihan, G. E.-H. (2006) Two randomized vitamin D trials in ambulatory patients on anticonvulsants Impact on bone. *Neurology,* **67**(11), 2005-2014.

Milberger, S., LeRoy, B., Lachance, L. & Edelson, G. (2002) Osteoporosis and women with Down syndrome. *Journal of Intellectual and Developmental Disability,* **27**(4), 273-280.

Milbrandt, T. A. & Johnston, C. E. (2005) Down syndrome and scoliosis: a review of a 50-year experience at one institution. *Spine,* **30**(18), 2051-2055.

Miller, B., Fridline, M., Liu, P.-Y. & Marino, D. (2014) Use of CHAID decision trees to formulate pathways for the early detection of metabolic syndrome in young adults. *Computational and mathematical methods in medicine,* **2014**.

Moher, D., Altman, D. G., Liberati, A. & Tetzlaff, J. (2011) PRISMA statement. *Epidemiology,* **22**(1), 128.

Morad, M., Nelson, N. P., Merrick, J., Davidson, P. W. & Carmeli, E. (2007) Prevalence and risk factors of constipation in adults with intellectual disability in residential care centers in Israel. *Research in Developmental Disabilities,* **28**(6), 580-6.

Murphy, E. L. & Comiskey, C. M. (2013) Using chi-Squared Automatic Interaction Detection (CHAID) modelling to identify groups of methadone treatment clients experiencing significantly poorer treatment outcomes. *Journal of substance abuse treatment,* **45**(4), 343-349.

Murray, C. J., Vos, T., Lozano, R., Naghavi, M., Flaxman, A. D., Michaud, C., Ezzati, M., Shibuya, K., Salomon, J. A. & Abdalla, S. (2013) Disability-adjusted life years (DALYs) for 291 diseases and injuries in 21 regions, 1990–2010: a systematic analysis for the Global Burden of Disease Study 2010. *The Lancet,* **380**(9859), 2197-2223.

National Osteoporosis Foundation (2010) *Clinician's Guide to Prevention and Treatment of Osteoporosis, Accessed: http://nof.org/files/nof/public/content/file/344/upload/159.pdf April 2013,* National Osteoporosis Foundation, Washington, DC.

Nguyen, T. V., Center, J. R. & Eisman, J. A. (2004) Osteoporosis: underrated, underdiagnosed and undertreated. *Medical journal of Australia,* **180**(5), S18.

Njeh, C., Boivin, C. & Langton, C. (1997) The role of ultrasound in the assessment of osteoporosis: a review. *Osteoporosis International,* **7**(1), 7-22.

Nocon, M., Hiemann, T., Müller-Riemenschneider, F., Thalau, F., Roll, S. & Willich, S. N. (2008) Association of physical activity with all-cause and cardiovascular mortality: a systematic review and meta-analysis. *European Journal of Cardiovascular Prevention & Rehabilitation,* **15**(3), 239-246.

Nolan, A., O'Regan, C., Dooley, C., Wallace, D., Hever, A., Cronin, H., Hudson, E. & Kenny, R. A. (2014) The Over 50s in a Changing Ireland Economic Circumstances, Health and Well-Being. Trinity College Dublin, Dublin.

Normand, C., Kamiya, Y., Timonen, V. & Whelan, B. (2011) Health and Social Care Utilisation. IN: *Fifty plus in Ireland 2011: first results from the Irish longitudinal study on ageing (TILDA).* (eds A. Barrett, G. Savva, V. Timonen & R. A. Kenny). Trinity College Dublin, Dublin.

O'Connell, M. B., Madden, D. M., Murray, A. M., Heaney, R. P. & Kerzner, L. J. (2005) Effects of proton pump inhibitors on calcium carbonate absorption in women: a randomized crossover trial. *The American journal of medicine,* **118**(7), 778-781.

O'Regan, C., Cronin, H. & Kenny, R. A. (2011) Mental Health and Cognitive Function IN: *Fifty Plus in Ireland 2011; First results from the Irish Longitudinal Study on Ageing (TILDA),*(eds Barrett, A, Savva, G,Timonen V and Kenny RA.). 157-180.

Odén, A., McCloskey, E., Kanis, J., Harvey, N. & Johansson, H. (2015) Burden of high fracture probability worldwide: secular increases 2010–2040. *Osteoporosis International,* **26**(9):2243-8.

O'Dwyer, M (2015) Prevalence, patterns and factors associated with multiple medicines use in an ageing population with intellectual disability in Ireland (Unpublished thesis).

O'Dwyer, M., Peklar, J., McCarron, M., McCallion, P. & Henman, M. (2013) Prevalence, Patterns and Factors Associated with Polypharmacy and Excessive Polypharmacy in an Ageing Population with Intellectual Disability in Ireland. In: *Irish Journal of Medical Science.* pp. S210-S210. Springer London Ltd 236 Grays Inn Rd, 6TH Floor, London WC1X 8HL, England.

Oliver, M. (1992) Changing the social relations of research production? *Disability, Handicap & Society,* **7**(2), 101-114.

Oliver, M. (2002) Emancipatory research: a vehicle for social transformation or policy development. In: *Inaugural NDA Disability Research Conference, Dublin, 3 December. Conference Proceedings: Using Emancipatory Methodologies in Disability Research.* pp. 15-23.

Ouellette-Kuntz, H. (2005) Understanding health disparities and inequities faced by individuals with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities,* **18**(2), 113-121.

Pallant, J. (2013) *SPSS survival manual,* McGraw-Hill Education (UK).

Parker, S. E., Troisi, R., Wise, L.A., Palmer, J.R., Titus-Ernstoff, L., Strohsnitter, W.C., & Hatch, E. E. (2013). Menarche, menopause, years of menstruation, and the incidence of osteoporosis: the influence of prenatal exposure to diethylstilbestrol. *The Journal of Clinical Endocrinology & Metabolism,* **99**(2), 594-601.

Pearson, D. A. (2007) Bone health and osteoporosis: the role of vitamin K and potential antagonism by anticoagulants. *Nutrition in Clinical Practice,* **22**(5), 517-544.

Peklar, J., Henman, M. C., Richardson, K., Kos, M. & Kenny, R. A. (2013) Food supplement use in the community dwelling population aged 50 and over in the Republic of Ireland. *Complementary therapies in medicine,* **21**(4), 333-341.

Penedo, F.J., & Dahn, J.R. (2005). Exercise and well-being: a review of mental and physical health benefits associated with physical activity. *Current Opinion in Psychiatry,* **18**(2), 189-193.

Perry, J., Kerr, M., Felce, D., Bartley, S. & Tomlinson, J. (2008) Monitoring the public health impact of health checks for adults with a learning disability in Wales. *Newport,* **560**(240), 43.

Petrone, L. R. (2012) Osteoporosis in Adults with Intellectual Disabilities. *Southern medical journal,* **105**(2), 87-92.

Piachaud, J., Rohde, J., & Pasupathy, A. (1998). Health screening for people with Down's syndrome. *Journal of Intellectual Disability Research,* **42**(5), 341-345.

Podsiadlo, D. & Richardson, S. (1991) The timed" Up & Go": a test of basic functional mobility for frail elderly persons. *Journal of the American geriatrics Society,* **39**(2), 142.

Polit, D. F. & Beck, C. T. (2008) *Nursing research: Generating and assessing evidence for nursing practice,* Lippincott Williams & Wilkins.

Poulsen, A., Christensen, S., McLaughlin, J., Thomsen, R. W., Sørensen, H. T., Olsen, J. & Friis, S. (2009) Proton pump inhibitors and risk of gastric cancer: a population-based cohort study. *British journal of cancer,* **100**(9), 1503-1507.

Prasher, V. (1999). Down syndrome and thyroid disorders: a review. *Down Syndrome Research and Practice,* **6**(1), 25-42.

Puffer, S., Torgerson, D. J., Sykes, D., Brown, P. & Cooper, C. (2004) Health care costs of women with symptomatic vertebral fractures. *Bone,* **35**(2), 383-386.

Qiu, C., Chen, H., Wen, J., Zhu, P., Lin, F., Huang, B., Wu, P., Lin, Q., Lin, Y. & Rao, H. (2013) Associations between age at menarche and menopause with cardiovascular disease, diabetes, and osteoporosis in Chinese women. *The Journal of Clinical Endocrinology & Metabolism,* **98**(4), 1612-1621.

R Development Core Team (2011) A language and environment for statistical computing. R Project for Statistical Computing Vienna, Austria. http://www.r-project.org.

Ray, J. G., Papaioannou, A., Ioannidis, G. & Adachi, J. D. (2002) Anticonvulsant drug use and low bone mass in adults with neurodevelopmental disorders. *QJM: An International Journal of Medicine,* **95**(4), 219-223.

Richardson, W. S., Wilson, M. C., Nishikawa, J. & Hayward, R. S. (1995) The well-built clinical question: a key to evidence-based decisions. *Acp j club,* **123**(3), A12-3.

Rimmer, J. H. & Yamaki, K. (2006) Obesity and intellectual disability. *Mental retardation and developmental disabilities research reviews,* **12**(1), 22.

Rimmer, J. H. (2011) Promoting inclusive community-based obesity prevention programs for children and adolescents with disabilities: the why and how. *Childhood Obesity (Formerly Obesity and Weight Management),* **7**(3), 177-184.

Rimmer, J. H., Rowland, J. L. & Yamaki, K. (2007) Obesity and secondary conditions in adolescents with disabilities: Addressing the needs of an underserved population. *Journal of Adolescent Health,* **41**(3), 224-229.

Rimmer, J., Yamaki, K., Lowry, B., Wang, E. & Vogel, L. (2010) Obesity and obesity-related secondary conditions in adolescents with intellectual/developmental disabilities. *Journal of Intellectual Disability Research,* **54**(9), 787-794.

Robertson, J., Emerson, E., Gregory, N., Hatton, C., Turner, S., Kessissoglou, S. & Hallam, A. (2000) Lifestyle related risk factors for poor health in residential settings for people with intellectual disabilities. *Research in developmental disabilities,* **21**(6), 469-486.

Robertson, J., Roberts, H., Emerson, E., Turner, S. & Greig, R. (2011) The impact of health checks for people with intellectual disabilities: a systematic review of evidence. *Journal of Intellectual Disability Research,* **55**(11), 1009-1019.

Rödström, K., Bengtsson, C., Milsom, I., Lissner, L., Sundh, V. & Bjoürkelund, C. (2003) Evidence for a secular trend in menopausal age: a population study of women in Gothenburg. *Menopause,* **10**(6), 538-543.

Roizen, N. J., Magyar, C. I., Kuschner, E. S., Sulkes, S. B., Druschel, C., van Wijngaarden, E., Rodgers, L., Diehl, A., Lowry, R. & Hyman, S. L. (2014) A community cross-sectional survey of medical problems in 440 children with Down syndrome in New York State. *The Journal of pediatrics,* **164**(4), 871-875.

Roth, G. M., Sun, B., Greensite, F. S., Lott, I. T., & Dietrich, R.B. (1996). Premature aging in persons with Down syndrome: MR findings. *American Journal of Neuroradiology,* **17**(7), 1283-1289.

Rubin, S.S., Rimmer, J.H., Chicoine, B., Braddock, D., & McGuire, D.E. (1998). Overweight prevalence in persons with Down syndrome. *Mental Retardation,* **36**(3), 175-181.

Sakadamis, A., Angelopoulou, N., Matziari, C., Papameletiou, V. & Souftas, V. (2002) Bone mass, gonadal function and biochemical assessment in young men with trisomy 21. *Eur J Obstet Gynecol Reprod Biol,* **100**(2), 208-12.

Sambrook, P. N., Cameron, I. D., Chen, J. S., Cumming, R. G., Lord, S. R., March, L. M., Schwarz, J., Seibel, M. J. & Simpson, J. M. (2007) Influence of fall related factors and bone strength on fracture risk in the frail elderly. *Osteoporosis International: A Journal Established As Result Of Cooperation Between The European Foundation For Osteoporosis And The National Osteoporosis Foundation Of The USA,* **18,** 603-610.

Samuel, M. J. (2015) American Geriatrics Society 2015 Updated Beers Criteria for Potentially Inappropriate Medication Use in Older Adults. *Journal of the American Geriatrics Society,* **63**(11), 2227-2246.

Sato, Y., Kondo, I., Ishida, S., Motooka, H., Takayama, K., Tomita, Y., Maeda, H. & Satoh, K. (2001) Decreased bone mass and increased bone turnover with valproate therapy in adults with epilepsy. *Neurology,* **57**(3), 445-449.

Schalock, R. L., Borthwick-Duffy, S. A., Bradley, V. J., Buntinx, W. H., Coulter, D. L., Craig, E. M., Gomez, S. C., Lachapelle, Y., Luckasson, R. & Reeve, A. (2010) *Intellectual disability: Definition, classification, and systems of supports,* ERIC.

Schalock, R., Borthwick-Duffy, S., Bradley, V., Buntinx, W. H., Coulter, D. L., Craig, E. M. P., Gomez, S. C., Lachapelle, Y., Luckasson, R., Reeve, A., Shogren, K. A., Snell, M. E., Spreat, S., Tassé, M. J., Thompson, J. R., Verdugo-Alonso, M. A., Wehmeyer, M. L. & Mark H Yeager (2010) Intellectual Disability: Definition, Classification, and Systems of Supports. American Association of Intellectual and Developmental Disabilities Washington, D.C.

Schmidt, E. V., Byars, J. R., Flamuth, D. H., Schott, J. J. & Sever, C. M. (2004) Prevalence of low bone-mineral density among mentally retarded and developmentally disabled residents in intermediate care. *Consult Pharm,* **19**(1), 45-51.

Schrager, S., Kloss, C. & Ju, A. W. (2007) Prevalence of fractures in women with intellectual disabilities: a chart review. *Journal of Intellectual Disability Research,* **51**(4), 253-259.

Schupf, N., Zigman, W., Kapell, D., Lee, J.H., Kline, J., & Levin, B. (1997). Early menopause in women with Down's syndrome. *Journal of Intellectual Disability Research,* **41**(3), 264-267.

Schwab, P., & Scalapino, K. (2011). Exercise for bone health: rationale and prescription. *Current Opinion in Rheumatology,* **23**(2), 137-141.

Seeman, E. & Delmas, P. D. (2006) Bone quality—the material and structural basis of bone strength and fragility. *New England Journal of Medicine,* **354**(21), 2250-2261.

Seeman, E. (1999). The structural basis of bone fragility in men. *Bone,* **25**(1), 143-147.

Seeman, E. (2002) Pathogenesis of bone fragility in women and men. *The Lancet,* **359**(9320), 1841-1850.

Seeman, E., Melton III, L. J., O'Fallon, W. M. & Riggs, B. L. (1983) Risk factors for spinal osteoporosis in men. *The American journal of medicine,* **75**(6), 977-983.

Sepúlveda, D., Allison, D.B., Gomez, J. E., & Kreibich, K. (1995). Low spinal and pelvic bone mineral density among individuals with Down syndrome. *American Journal on Mental Retardation,* **100**(2), 109-114.

Singh, K. & Xie, M. (2008) Bootstrap: A Statistical Method. Accessed: October 2015 http://stat.rutgers.edu/home/mxie/RCPapers/bootstrap.pdf Rutgers University

Sinnema, M., Maaskant, M. A., van Schrojenstein Lantman-de Valk, H. M., van Nieuwpoort, I. C., Drent, M. L., Curfs, L. M. & Schrander-Stumpel, C. T. (2011) Physical health problems in adults with Prader-Willi syndrome. *Am J Med Genet A,* **155A**(9), 2112-24.

Siris, E. S., Boonen, S., Mitchell, P. J., Bilezikian, J. & Silverman, S. (2012) What's in a name? What constitutes the clinical diagnosis of osteoporosis? *Osteoporos Int,* **23**(8), 2093-7.

Siris, E. S., Harris, S. T., Rosen, C. J., Barr, C. E., Arvesen, J. N., Abbott, T. A. & Silverman, S. (2006) Adherence to bisphosphonate therapy and fracture rates in osteoporotic women: relationship to vertebral and nonvertebral fractures from 2 US claims databases. *Mayo Clin Proc,* **81**(8), 1013-22.

Siris, E. S., Miller, P. D., Barrett-Connor, E., Faulkner, K. G., Wehren, L. E., Abbott, T. A., Berger, M. L., Santora, A. C. & Sherwood, L. M. (2001) Identification and fracture outcomes of undiagnosed low bone mineral density in postmenopausal women: results from the National Osteoporosis Risk Assessment. *JAMA,* **286**(22), 2815-22.

Sowney, M. & Barr, O. (2004) Equity of Access to Health Care for People with Learning Disabilities A concept analysis. *Journal of Learning Disabilities,* **8**(3), 247-265.

Srikanth, R., Cassidy, G., Joiner, C., & Teeluckdharry, S. (2011). Osteoporosis in people with intellectual disabilities: A review and a brief study of risk factors for osteoporosis in a community sample of people with intellectual disabilities. *Journal of Intellectual Disability Research,* **55**(1), 53-62. doi: 10.1111/j.1365-2788.2010.01346.x

Stevenson, R. D., Conaway, M., Chumlea, W. C., Rosenbaum, P., Fung, E. B., Henderson, R. C., Worley, G., Liptak, G., O'Donnell, M. & Samson-Fang, L. (2006) Growth and health in children with moderate-to-severe cerebral palsy. *Pediatrics,* **118**(3), 1010-1018.

Tabachnick, B. G., Fidell, L. S. & Osterlind, S. J. (2001) Using multivariate statistics. Pearsons, Boston.

Taggart, L. & Cousins, W. (2014) *Health promotion for people with intellectual and developmental disabilities,* McGraw-Hill Education (UK).

Tang, B.M.P., Eslick, G.D., Nowson, C., Smith, C., & Bensoussan, A. (2007). Use of calcium or calcium in combination with vitamin D supplementation to prevent fractures and bone loss in people aged 50 years and older: a meta-analysis. *The Lancet,* **370**(9588), 657-666.

Targownik, L. E., Lix, L. M., Metge, C. J., Prior, H. J., Leung, S. & Leslie, W. D. (2008) Use of proton pump inhibitors and risk of osteoporosis-related fractures. *Canadian Medical Association Journal,* **179**(4), 319-326.

Taylor, J. L. & Hodapp, R. M. (2012) Doing Nothing: Adults With Disabilities With No Daily Activities and Their Siblings. *American Journal on Intellectual and Developmental Disabilities,* **117**(1), 67-79.

Tohill, C. & Laverty, A. (2001) Sunshine, diet and mobility for healthy bones -- an intervention study designed to implement these standards into the daily routine in an at risk population of adults with intellectual disability. *Journal of Intellectual & Developmental Disability,* **26**(3), 217-231.

Torr, J., Strydom, A., Patti, P., & Jokinen, N. (2010). Aging in Down Syndrome: Morbidity and Mortality. *Journal of Policy and Practice in Intellectual Disabilities,* **7**(1), 70-81. doi: 10.1111/j.1741-1130.2010.00249.x

Tracy, J. & McDonald, R. (2015) Health and Disability: Partnerships in Health care. *Journal of Applied Research in Intellectual Disabilities,* **28**(1), 22-32.

Tsimaras, V., Giagazoglou, P., Fotiadou, E., Christoulas, K., & Angelopoulou, N. (2003). Jog-walk training in cardiorespiratory fitness of adults with Down syndrome. *Perceptual & Motor Skills,* **96**(3), 1239.

Turner, S. & Robinson, C. (2011) Reasonable Adjustments for People with Learning Disabilities-Implications and Actions for Commissioners and Providers of Healthcare. *Improving Health and Lives.*

Tyler, C. V., Jr., Snyder, C. W. & Zyzanski, S. (2000) Screening for osteoporosis in community-dwelling adults with mental retardation. *Ment Retard,* **38**(4), 316-21.

Tyrrell, J., Cosgrave, M., McCarron, M., McPherson, J., Calvert, J., Kelly, A., McLaughlin, M., Gill, M. & Lawlor, B. (2001) Dementia in people with Down's syndrome. *International Journal of Geriatric Psychiatry,* **16**(12), 1168-1174.

Väänänen, H. K. & Zhao, H. (2008) Osteoclast function: biology and mechanisms. *Principles of Bone Biology, 3rd ed. Academic Press, San Diego, USA,* 193-209.

van Allen, M.I., Fung, J., & Jurenka, S.B. (1999). Health care concerns and guidelines for adults with Down syndrome. *American Journal of Medical Genetics,* **89**(2), 100-110.

van Schrojenstein Lantman-De, H. M., Metsemakers, J. F., Haveman, M. J. & Crebolder, H. F. (2000) Health problems in people with intellectual disability in general practice: a comparative study. *Family practice,* **17**(5), 405-407.

van Schrojenstein Lantman-deValk, H., Schupf, N. & Patja, K. (2002) Reproductive and physical health. *Health of women with intellectual disabilities,* 22-40.

Vanlint, S. & Nugent, M. (2006) Vitamin D and fractures in people with intellectual disability. *J Intellect Disabil Res,* **50**(10), 761-7.

Vestergaard, P., Prieto-Alhambra, D., Javaid, M. K. & Cooper, C. (2013) Fractures in users of antidepressants and anxiolytics and sedatives: effects of age and dose. *Osteoporosis International,* **24**(2), 671-680.

Vestergaard, P., Rejnmark, L. & Mosekilde, L. (2005) Relative fracture risk in patients with diabetes mellitus, and the impact of insulin and oral antidiabetic medication on relative fracture risk. *Diabetologia,* **48**(7), 1292-1299.

Vice, M. A., Nahar, V. K., Ford, M. A., Bass, M. A., Johnson, A. K., Davis, A. B. & Biviji-Sharma, R. (2015) Risk Factors for Low Bone Mineral Density in Institutionalized Individuals with Developmental Disabilities. *Health promotion perspectives,* **5**(2), 147.

Von Bogdandy, A. (2000) The European Union as a human rights organization? Human rights and the core of the European Union. *Common Market Law Review,* **37,** 1307-1338.

Wade, D. & Collin, C. (1988) The Barthel ADL Index: a standard measure of physical disability? *International disability studies,* **10**(2), 64-67.

Wagemans, A., Fiolet, J., Van Der Linden, E. & Menheere, P. (1998) Osteoporosis and intellectual disability: is there any relation? *Journal of Intellectual Disability Research,* **42**(5), 370-374.

Walsh, J., Henry, Y., Fatayerji, D. & Eastell, R. (2009) Lumbar spine peak bone mass and bone turnover in men and women: a longitudinal study. *Osteoporosis international,* **20**(3), 355-362.

Warburton, D.E.R., Gledhill, N., & Quinney, A. (2001). The effects of changes in musculoskeletal fitness on health. *Canadian Journal of Applied Physiology,* **26**(2), 161-216.

Warburton, D.E.R., Nicol, C.W., & Bredin, S. S.D. (2006). Health benefits of physical activity: the evidence. *Canadian medical association journal,* **174**(6), 801-809.

Waung, J.A., Bassett, J.H., & Williams, G.R. (2012). Thyroid hormone metabolism in skeletal development and adult bone maintenance. *Trends in Endocrinology & Metabolism,* **23**(4), 155-162.

Weber, P. (2001) Vitamin K and bone health. *Nutrition,* **17,** 880-887.

Wentworth, J. (2012). Obesity and diabetes. *Diabetes Management: A Journal for General Practitioners and Other Health Professionals,* **39**, 26.

Wilkinson, J. E., Culpepper, L. & Cerreto, M. (2007) Screening tests for adults with intellectual disabilities. *Journal of the American Board of Family Medicine,* **20,** 399-407.

Wongdee, K. & Charoenphandhu, N. (2015) Update on type 2 diabetes-related osteoporosis. *World Journal of Diabetes,* **6**(5), 673.

World Health Organisation (1995) Physical status: the use and interpretation of anthropometry. Report of a WHO Expert Committee In: *WHO Technical Report Series 854.* Geneva: World Health Organization.

World Health Organisation (2011a) **WHO**. Anatomical Chemical Therapeutic Classification System http://www.whocc.no/atc_ddd_index/ accessed July 2011

World Health Organisation (2011b) Waist circumference and waist-hip ratio, Report of a WHO Expert Consultation Geneva, 8–11 December 2008 WHO Document Production Services, Geneva, Switzerland.

World Health Organisation (2015a) Regional Office for Europe; Obesity Data and Statistics, Accessed; http://www.euro.who.int/en/health-topics/noncommunicable-diseases/obesity/data-and-statistics April 2015. WHO, Geneva.

World Health Organization (1994). Assessment of fracture risk and its application to screening for postmenopausal osteoporosis: report of a WHO study group [meeting held in Rome from 22 to 25 June 1992]. Report of a WHO Study Group. *World Health Organ Tech Rep Ser,* **843,** 1-129.Accessed at: https://extranet.who.int/iris/restricted/bitstream /10665/39142/1/WHO_TRS_843_eng.pdf

World Health Organization (2002) Active ageing: A policy framework. Accessed: http://apps.who.int/iris/bitstream/10665/67215/1/WHO_NMH_NPH_02.8.pdf . March 2014. World Health Organisation Geneva.

World Health Organization (2015b) A global brief on hypertension: silent killer, global public health crisis. *Women.* Accessed: http://apps.who.int/iris/bitstream/10665/79059/1/WHO_DCO_WHD_2013.2_eng.pdf. December 2015.

World Health Organization. (2003). *Prevention and management of osteoporosis: report of a WHO scientific group*: World Health Organization. Diamond Pocket Books (P) Ltd. World Health Organization, Geneva Accessed at: http://apps.who.int/iris/bitstream /10665/42841/1/WHO_TRS_921.pdf

World Health Organization (2004) WHO scientific group on the assessment of osteoporosis at primary health care level. In: *Summary meeting report.* pp. 5-7 Accessed: http://www.who.int/chp/topics/Osteoporosis.pdf. June 2014.

World Health Organization. (2010). Global recommendations on physical activity for health. Geneva, Switzerland: World Health Organisation. Accessed at: http://apps.who.int/iris/bitstream/10665/44399/1/9789241599979_eng.pdf

Wortsman, J., Matsuoka, L.Y., Chen, T.C., Lu, Z., & Holick, M.F. (2000). Decreased bioavailability of vitamin D in obesity. *The American Journal of Clinical Nutrition,* **72**(3), 690-693.

Yamaki, K. (2005) Body weight status among adults with intellectual disability in the community. *Journal Information,* **43**(1).

Zimmerman. S., Hawkes, W., Chandler, J., Custis, B. V., Hebel, J., Sloanes, P., Martin, A., Magaziner, J., Holder, L. & Girman, C. (1998) Correlates of bone mineral density in nursing home residents. *The journal of nutrition, health & aging,* **3,** 140-145.

Zimmerman, S. I., Girman, C., Buie, V. C., Chandler, J., Hawkes, W., Martin, A., Holder, L., Hebel, J., Sloane, P. & Magaziner, J. (1999) The prevalence of osteoporosis in nursing home residents. *Osteoporosis International,* **9**(2), 151-157.

Zylstra, R. G., Porter, L. L., Shapiro, J. L. & Prater, C. D. (2008) Prevalence of osteoporosis in community-dwelling individuals with intellectual and/or developmental disabilities. *Journal of the American Medical Directors Association,* **9**(2), 109-113.

APPENDICES

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Appendix 1:

Systematic Review Protocol

OSTEOPOROSIS AND THE OLDER ADULT WITH INTELLECTUAL DISABILITY



Protocol for a systematic literature review

A systematic review protocol for the review of osteoporosis and osteopenia among older adults with an intellectual disability in Ireland.

Eilish Burke

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1.0 Title

Protocol for a systematic literature search of the prevalence of osteoporosis among older adults with intellectual disability

2.0 Definition of terms

2.1 Osteoporosis/Osteopenia: A multifactorial disorder characterised by low bone mass and micro-architectural deterioration of bone tissue leading to enhanced bone fragility and a consequent increase in fracture risk.

(Kanis *et al.* 2013, Siris *et al.* 2012, Mergler *et al.* 2009, Srikanth *et al.* 2011)

Previously osteoporosis was defined by the WHO (1994) provided diagnostic criteria for osteoporosis based on bone mineral density (BMD- t-score <-2.5 SD below the young adult mean), however the consensus now is that these parameters do not adequately identify those at risk of the consequence of osteoporosis, namely fracture, therefore the inclusion of risk factors is essential.

> A systematic evidence review is "a scientific investigation that focuses on a specific question and uses explicit, prespecified scientific methods to identify, select, assess, and summarize the findings of similar but separate studies. It may include a quantitative synthesis (meta-analysis), depending on the available data."
>
> The Institute of Medicine (IOM) 2011

2.2 Intellectual disability: An intellectual disability is a disability that involves significant limitations both in intellectual functioning and in adaptive behaviour, which covers many everyday social and practical skills. This disability originates before the age of 18 and comprises a wide range of conditions, types, and levels. Intellectual disability is caused by factors that can be physical, genetic, and/or social.

(Schalock *et al.* 2010)

Within this study the internationally recognised terms mild, moderate, severe and profound ID are used as descriptive terms only to distinguish a person level of ID.

2.3 Densitometry: Is an umbrella term adopted in this study to describe the methods used to examine bone quality and density.

2.4 DXA: The gold standard is the dual-energy x-ray absorptiometry (DXA) also known as Bone density scanning, or bone *densitometry*. This is an enhanced form of x-ray technology that is used to measure bone loss. DXA is today's established standard for measuring bone mineral density (BMD).

2.5 QUS: A surrogate measure of bone quality, quantitative ultrasound is a portable scan of the heel (calcaneus bone) to establish bone status. It is inexpensive, radiation free and provides a diagnostic alternative to DXA scanning.

3.0 Contextualising the Study Relevance

The prevalence of osteoporosis and osteopenia is known to increase with age (Siris *et al.* 2006) and subsequently there is an increased risk of fracture especially hip fractures which have been reported to occur predominantly over the age of 50 years (Sambrook *et al.* 2007). Kanis (2002) also note that age is associated with an increased risk of fracture independent of bone mineral density (BMD) possibly due to increased falls as a result of weaker muscles and poorer balance. The Irish Longitudinal Study on Ageing (TILDA) reported an increase in the prevalence of hip fracture with age from 2.8% (50-65 years) to 6.7% (75 years+) contributing to this increase in fracture risk is osteoporosis(Barrett *et al.* 2011). In Ireland it is estimated that over 300,000 people have osteoporosis with a steady increase predicted primarily due to an ageing population (Health Service Executive (HSE) 2008). It is estimated that one in three women and one in five men may have osteoporosis (HSE 2008). TILDA reported the prevalence of osteoporosis at 16.1% in females and 1.9% in males with a definitive age gradient (Cronin *et al.* 2011). However what is striking with this study is that the participants were also objectively measured for the presence of osteoporosis and there was a notable difference between the reported status and objectively measured status. Of those women who had an objective measurement of osteoporosis just 34% had reported a doctor's diagnosis and what was more alarming was that 100% of the men who were objectively measured with evidence of osteoporosis, did not report a doctor's diagnosis (Cronin *et al.* 2011). This emphasises the insidious nature of the disease which often goes unrecognised and untreated until a fracture occurs (Kanis *et al.* 2009). The resulting personal health burden and economic cost is considerable (Barrett-Connor 1995, Puffer *et al.* 2004) and the International Osteoporosis Foundation predicts this will continue to increase substantially into the future.

3.1 Bone health and people with ID

It is reported that people with ID present with increased risk of low bone mineral density (Aspray *et al.* 1998, Center *et al.* 1998, Baptista *et al.* 2005, Jaffe *et al.* 2005) and that osteoporosis continues to be under diagnosed and undertreated in the older population (Aspray et al. 1998, Jasien *et al.* 2012). International studies suggest that people with ID also present with a greater risk for poor bone health (Haveman *et al.* 2011) and in fact it is noted that people with ID and especially Down syndrome (DS) can have poorer health than the general population however are more likely to have infrequent health checks or routine screening (Michael 2008). Srikanth et al. (2011) report that one area often neglected in physical health checks for people with ID is bone health status. The Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing (IDS-TILDA) is the first large scale longitudinal study on people with ID investigating the key determinants of health and well-being in this population (McCarron *et al.* 2009) and provides the platform from which bone health and its impact on people's lives can be investigated. It is therefore prudent to examine the bone health of people with ID in Ireland at this time.

4.0 Developing a search strategy

The subject under consideration is the bone health of people with ID. There is a paucity of information on bone health and identifying and establishing valid data on the prevalence of poor bone health among people with ID will be important to clinical practice, policy development and for economic reasons.

4.1 Framing the question

The PICO model was used to develop the main concepts and formulate the question

Population: Adults with ID

Indicator: Osteoporosis, low t-score, bone mineral density, history of fracture, identification of specific risk factors, have had a DXA or QUS.

Comparative: not applicable as prevalence only is being identified.

Outcome: Prevalence of poor bone health

Question:

What is the prevalence of osteoporosis/osteopenia among adults with intellectual disability?

5.0 Methods

All studies reporting prevalence of osteoporosis or osteopenia among people with an intellectual disability will be included in this review. To guide the identification of the search inclusion and exclusion criteria will be applied. A four phase flow as described by Liberati et al (2009) will be utilised to guide the extraction of appropriate literature. This involves

- Identification
- Screening
- Eligibility
- Including

5.1 Identification

The following eligibility criteria and exclusion criteria will apply.

5.1.1 Eligibility Criteria

Types of studies

Primary studies, peer reviewed, includes adults only with an intellectual disability, English language

Study designs: Cross-sectional, cohort studies, RTC.

Types of participants: Adults 18 years + with intellectual disability will be considered

Types of outcome measures

Identification of the prevalence of osteoporosis/osteopenia from bone scanning

- DXA
- QUS

Identify and name where possible the types of devices used as they differ in how their measurements are expressed

5.1.2 Exclusion Criteria

The following criteria will be considered when excluding articles from the literature search.

- Osteoporosis or osteopenia prevalence in the general population
- Children with or without ID
- Specific syndrome synonymous with osteoporosis or low BMD – Retts syndrome, PWS, as these results would effectively skew the results
- Case study design
- It is also proposed to exclude grey literature

The reference lists of all the RCTs and reviews will be manually searched to obtain any other possible eligible studies

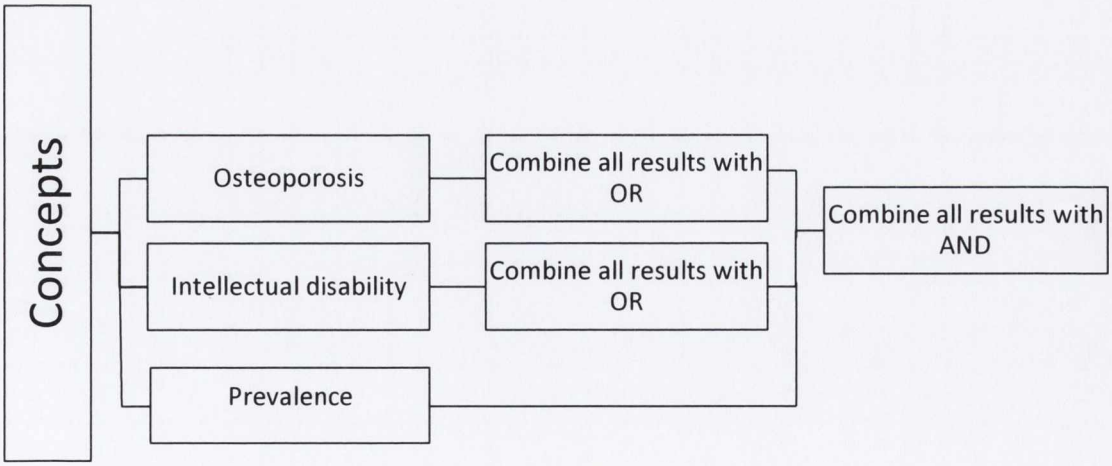
5.1.3 Databases identified

It is proposed to use the following 4 databases

- CINAHL
- psycINFO
- MEDLINE
- EMBASE

5.1.4 Article Identification

The following broad concepts have been identified as concepts that will address the research question. These concepts have been further subdivided into search terms which will be inputted into the identified databases. Each search term will be individually searched within the database and then the search terms from each concept will be combined with 'OR'. Following this the results from each of the concepts will be combined with 'AND'. The final results from each of the databases will then be screened. An independent second reviewer shall run the concept map in the identified databases and check for accuracy, quality of identified articles and match. Any literature not previously identified shall be included and screened as per protocol moving forward.



5.2 Article Screening

Following extensive searching of each of the databases, the results saved, all titles will be preliminary screened for applicability to inclusion criteria. Any papers with titles that pose ambiguity will be retained. This will result in a smaller number of applicable papers which will then be examined by abstract for applicability to inclusion criteria. Again any articles which pose ambiguity shall be retained. Full text papers of the final number of papers identified will be extracted, read and scrutinised for appropriateness and applicability to research question. An independent second reviewer

5.3 Articles Included

Any paper that violates the inclusion criteria will be excluded, careful notes of rationale for exclusion will be kept at all stages of the screening process. The final article number included will then be analysed, graded for quality and specific data extracted.

5.4 Quality Assurance

A number of quality assessment tools and procedures were considered for the quality assessment of bias in this systematic review. For example Newcastle-Ottawa Scale (NOS); Steps in Conduction Methodology Quality, Fink, 2005; AHRQ, Methods Guide for Effectiveness and Comparative Effectiveness Reviews; Scottish SIGN and Cochrane Handbook. Based on their applicability, user friendliness and clarity and in consultation with the independent second reviewer the National Institute of Health guidelines for assessing quality of case-Control Studies, Observational and Cross-sectional studies were chosen. The methodological quality assurance procedure from the National Institute of Health (US, National health and human services) can be retrieved from http://www.nhlbi.nih.gov/health-pro/guidelines/in-develop/cardiovascular-risk-reduction/tools/.

This will evaluate the study's research design, data collection, data analysis and results and methodological limitations. The guidelines are divided into assessing 'case-control studies' and 'observation or cross sectional studies'. The assessor identifies whether or not the article meets the criteria and based on the overall score out of, 12 quality probing questions in the case of 'case-control studies' and out of 14 otherwise, the overall quality is assessed and rated into three categories, good, fair and poor. When 'no' or other rating is applied the assessor must consider the risk of bias that could be introduced by that flaw to the review and if article is then overall ranked as 'poor' rationale for this rating must be included in the evaluation form. The rating guideline score has been previously examined, discussed and decided by the assessor and her

independent second. See table 1 for description of quality assessment scores. On completion of quality assessment the remaining articles will then go forward for data extraction.

Table 1.0 Quality Assessment Scores

Rating	Case-Control Studies	Observational Cohort & Cross-Sectional Studies	Action
	Confirmed Yes	Confirmed Yes	
Good	9-12	10-14	Move to data extraction
Fair	6-8	7-9	Adjudicate with 3rd independent assessor if no higher quality study identified and consider borderline studies (i.e. those that scored 8 for case-control or 9 for observational cohort)
Poor	≤5	≤6	Reject
Other	CD, NR, NA*	CD, NR, NA*	Consider adjudication

*CD - Cannot determine; NR – Not applicable; NA – Not reported

6.0 Data extraction

Each article will be scrutinised and the following framework will be utilised to extract the necessary data (see table 2.0)

Table 2.0 Data extraction form

No	Author	Study design	Age	Population								Specific living circumstance				Type of equipment utilised to establish prevalence	Prevalence data	Comments
		Observational /Case control		Men only	Women only	Post-menopausal Women	Men and women	Level of ID	aetiology specific	Down syndrome	Other aetiology	Independent	Community group home	Residential or congregated	None specified		% (raw data)	

6.1 Analysis and synthesis

Give an overall description of the studies found identify their adherence to the inclusion criteria. Present all studies with summary in table format, method of diagnosis, prevalence identified and identify level of quality assigned to paper.

7.0 Discussion and conclusion

Discuss findings in light of current general literature on prevalence among general population. Draw conclusion on quality of evidence found, provide implications for research and practice.

Appendix 2.1

Systematic search strategy

Systematic Search Strategy:

Research Question: what is the prevalence of osteoporosis and osteopenia among older adults with intellectual disability?

Databases Selected: CINAHL; PubMed; PsychInfo; Embase:

Filters Applied: None

Concept 1: Osteoporosis

Pubmed MeSH Term: Osteoporosis OR "Osteoporosis, Postmenopausal"

Medline: (MH "Osteoporosis") OR (MH "Osteoporosis, Postmenopausal")

CINAHL: Osteoporosis

PsycInfo: DE "Osteoporosis"

Embase: 'osteoporosis'/de OR 'senile osteoporosis'/exp OR 'secondary osteoporosis'/exp OR 'primary osteoporosis'/exp OR 'postmenopause osteoporosis'/ex

Free Text: Osteoporoses OR "Osteoporosis, Post-Traumatic" OR "Osteoporosis, Post Traumatic" OR "Post-Traumatic Osteoporoses" OR "Post-Traumatic Osteoporosis" OR "Osteoporosis, Senile" OR "Osteoporoses, Senile" OR "Senile Osteoporoses" OR "Senile Osteoporosis" OR "Osteoporosis, Involutional" OR "Osteoporosis, Age-Related" OR "Osteoporosis, Age Related" OR "Bone Loss, Age-Related" OR "Age-Related Bone Loss" OR "Age-Related Bone Losses" OR "Bone Loss, Age Related" OR "Bone Losses, Age-Related" OR "Age-Related Osteoporosis" OR "Age Related Osteoporosis" OR "Age-Related Osteoporoses" OR "Osteoporoses, Age-Related" OR "Osteoporosis, Postmenopausal" OR Bone Loss, Postmenopausal OR "Bone Losses, Postmenopausal" OR "Postmenopausal Bone Losses" OR "Postmenopausal Osteoporosis" OR "Osteoporoses, Postmenopausal" OR "Postmenopausal Osteoporoses" OR "Perimenopausal Bone Loss" OR "Postmenopausal Bone Loss" OR "Bone Loss, Perimenopausal" OR "Bone Losses, Perimenopausal" OR "Perimenopausal Bone Losses" OR "Osteoporosis, Post-Menopausal" OR "Osteoporoses, Post-Menopausal" OR "Osteoporosis, Post Menopausal" OR "Post-Menopausal Osteoporoses" OR "Post-Menopausal Osteoporosis" OR Osteopenia OR Osteopenias OR "brittle bone" OR "brittle bones" OR "weak bone" OR "weak bones" OR "frail bone" OR "frail bones" OR "bone fragility" OR "bone strength" OR "bone depletion" OR "demineralised bone" OR "demineralized bone" OR "bone mineral density" OR "bone mineral content" OR "bone density" OR "bone strength" OR "bone remodelling"

<u>Concept 2:</u> Intellectual Disability

Pubmed: Intellectual Disability

MEDLINE: Intellectual Disability

CINAHL: Intellectual Disability

Embase: EMTREE 'intellectual impairment'/exp OR Down syndrome/exp

PsychInfo: (DE "Learning Disabilities" OR DE "Developmental Disabilities") OR (DE "Intellectual Development Disorder" OR DE "Down's Syndrome" OR DE "Cognitive Impairment" OR DE "Developmental Disabilities" OR DE "Specific Language Impairment")

Free Text: "Intellectual Disability" OR "Disabilities, Intellectual" OR "Intellectual Disabilities" OR "Mental Retardation" OR "Retardation, Mental" OR "Disability, Intellectual" OR "Mental Retardation, Psychosocial" OR "Mental Retardations, Psychosocial" OR "Psychosocial Mental Retardation" OR "Psychosocial Mental Retardations" OR "Retardation, Psychosocial Mental" OR "Retardations, Psychosocial Mental" OR "Deficiency, Mental" OR "Deficiencies, Mental" OR "Mental Deficiencies" OR "Mental Deficiency" OR Idiocy OR "cognitive impairment" OR "cognitive impairments" OR "learning disability" OR "learning disabilities" OR "mentally handicapped" OR "mental handicap" OR "Intellectual Development Disorder" OR "Intellectual Development Disorders" OR "intellectual impairment" OR "intellectual impairments"

<u>Prevalence:</u>

(Prevalence OR Prevalences OR Occurrence OR occurrences OR incidence OR incidences OR frequency OR instance OR instances OR occurred OR occurring OR existent OR extent)

Appendix 2.2

Quality Assessment of Case-Control Studies

Criteria	Yes	No	Other (CD, NR, NA)*
1. Was the research question or objective in this paper clearly stated and appropriate?			
2. Was the study population clearly specified and defined?			
3. Did the authors include a sample size justification?			
4. Were controls selected or recruited from the same or similar population that gave rise to the cases (including the same timeframe)?			
5. Were the definitions, inclusion and exclusion criteria, algorithms or processes used to identify or select cases and controls valid, reliable, and implemented consistently across all study participants?			
6. Were the cases clearly defined and differentiated from controls?			
7. If less than 100 percent of eligible cases and/or controls were selected for the study, were the cases and/or controls randomly selected from those eligible?			
8. Was there use of concurrent controls?			
9. Were the investigators able to confirm that the exposure/risk occurred prior to the development of the condition or event that defined a participant as a case?			
10. Were the measures of exposure/risk clearly defined, valid, reliable, and implemented consistently (including the same time period) across all study participants?			
11. Were the assessors of exposure/risk blinded to the case or control status of participants?			
12. Were key potential confounding variables measured and adjusted statistically in the analyses? If matching was used, did the investigators account for matching during study analysis?			
Quality Rating (Good, Fair, or Poor) (see guidance)			
Rater #1 initials:			

Rater #2 initials:
Additional Comments (If POOR, please state why):

*CD, cannot determine; NA, not applicable; NR, not reported

Appendix 2.3

Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies

Criteria	Yes	No	Other (CD, NR, NA)*
1. Was the research question or objective in this paper clearly stated?			
2. Was the study population clearly specified and defined?			
3. Was the participation rate of eligible persons at least 50%?			
4. Were all the subjects selected or recruited from the same or similar populations (including the same time period)? Were inclusion and exclusion criteria for being in the study prespecified and applied uniformly to all participants?			
5. Was a sample size justification, power description, or variance and effect estimates provided?			
6. For the analyses in this paper, were the exposure(s) of interest measured prior to the outcome(s) being measured?			
7. Was the timeframe sufficient so that one could reasonably expect to see an association between exposure and outcome if it existed?			
8. For exposures that can vary in amount or level, did the study examine different levels of the exposure as related to the outcome (e.g., categories of exposure, or exposure measured as continuous			

variable)?			
9. Were the exposure measures (independent variables) clearly defined, valid, reliable, and implemented consistently across all study participants?			
10. Was the exposure(s) assessed more than once over time?			
11. Were the outcome measures (dependent variables) clearly defined, valid, reliable, and implemented consistently across all study participants?			
12. Were the outcome assessors blinded to the exposure status of participants?			
13. Was loss to follow-up after baseline 20% or less?			
14. Were key potential confounding variables measured and adjusted statistically for their impact on the relationship between exposure(s) and outcome(s)?			
Quality Rating (Good, Fair, or Poor) (see guidance)			
Rater #1 initials:			
Rater #2 initials:			
Additional Comments (If POOR, please state why):			

*CD, cannot determine; NA, not applicable; NR, not reported

Guidance for Assessing the Quality of Case-control, Observational Cohort and Cross-Sectional Studies may be accessed at http://www.nhlbi.nih.gov/health-pro/guidelines/in-develop/cardiovascular-risk-reduction/tools/cohort.

Appendix 2:

Literature Search Decisions

Article selection and quality assessment

Articles chosen following screen by title and reference list scrutiny (N=50)

✓=Accepted-Going forward for quality check (QC) X=Rejected ?=Query unsure for deliberation FF=Fatal Flaw FFDE=Forward For Data Extraction

No.	Article	Result	Second review consult with independent reviewer	Rationale	Type of Study		Quality Ass. Result		
					Cohort	Case-control	EB	MOD	FFDE
1	Angelopoulou, N., Matziari, C., Tsimaras, V., Sakadamis, A., Souftas, V., & Mandroukas, K. (2000). Bone mineral density and muscle strength in young men with mental retardation (with and without Down syndrome). *Calcif Tissue Int, 66*(3), 176-180.	✓	✓	22 participants, 26 years average BMD – DXA Compared to 27 health/age matched		✓	9/12 Good	9/12 Good	😊
2	Angelopoulou, N., Souftas, V., Sakadamis, A., & Mandroukas, K. (1999). Bone mineral density in adults with Down's syndrome. *Eur Radiol, 9*(4), 648-651.	✓	✓	Men only DXA – mean BMD 3 groups DS/ID/Control (8,8,10; N=26)		✓	9/12 Good	9/12 Good	😊
3	Arvio, M., Kilpinen-Loisa, P., Tiitinen, A., Huovinen, K., & Mäkitie, O. (2009). Bone mineral density and sex hormone status in intellectually disabled women on progestin-induced amenorrhea. *Acta Obstetricia Et Gynecologica Scandinavica, 88*(4), 428-433. doi: 10.1080/00016340902763244	✓	✓	Women only N=51 Avg. age 45 controls 115 (staff)			8/12 Fair	8/12 Fair	😐
4	Aspray, T. J., Francis, R. M., Thompson, A., Quilliam, S. J., Rawlings, D. J., & Tyrer, S. P. (1998). Comparison of ultrasound measurements at the heel between adults with mental retardation and control subjects. *Bone, 22*(6), 665-668.	✓	✓	N=170 ID N=108 Controls – age and gender matched BMD – QUS cubba -		✓	7/12 Fair	7/12 Fair	😐
5	Baptista, F., Varela, A., & Sardinha, L. B. (2005). Bone mineral mass in males an females with and without Down syndrome. *Osteoporosis International, 16*(4), 380-388. doi: 10.1007/s00198-004-1687-1	?	X	Age range 14 – 40 (children) 33 female; 34 male IQ determined level of ID Compared to ge and gender matched control BMD – DXA 24 females DS 28 males DS	Did not meet criteria –included children in analysis				

6	Bastiaanse, L. P., Mergler, S., Evenhuis, H. M., & Echteld, M. A. (2014). Bone quality in older adults with intellectual disabilities. *Res Dev Disabil, 35*(9), 1927-1933. doi: 10.1016/j.ridd.2014.04.018	✓	✓	768 ID QUS –BMD – Lunar Achilles	✓		8/14 Fair	8/14 Fair	😐
7	Carfi, A., Antocicco, M., Brandi, V., Cipriani, C., Fiore, F., Mascia, D., . . . Onder, G. (2014). Characteristics of adults with down syndrome: prevalence of age-related conditions. *Front Med (Lausanne), 1*, 51. doi: 10.3389/fmed.2014.00051	X	X	Chart/GP review Osteoporosis was not objectively measured	Did not meet criteria – BMD determined by chart review and was not objectively measured for study				
8	Center, J., Beange, H., & McElduff, A. (1998). People with mental retardation have an increased prevalence of osteoporosis: a population study. *American Journal Of Mental Retardation: AJMR, 103*(1), 19-28.	✓	✓	N=94 DXA – lumbar spine	✓		10/14 Good	10/14 Good	😊
9	Center, J., McElduff, A., & Beange, H. (1994). Osteoporosis in groups with intellectual disability 1. *Journal of Intellectual and Developmental Disability, 19*(4), 251-258.	✓	✓	N=53 n=10 had BMD 43 had clinical assessment - 31 females/22 males	✓		5/14 Poor	5/14 Poor	😐
10	Chen, P. H., Chen, C. Y., Lin, Y. C., & Chen, M. Y. (2015). Low bone mineral density among adults with disabilities in Taiwan: A cross-sectional descriptive study. *Disability And Health Journal.*	?	✓	Study included all disabilities (physical and ID) Sample N=572 ID – n=211 BMD – QUS Lunar Achilles Groups results are mixed for PWID does not give osteoporosis and osteopenia separately – 55.7%	✓		7/14 Fair	7/14 Fair	😐
11	Curtis, R., Freitag, P., LaGuardia, J. J., Thornton, S., Vicari, S., & Markwell, S. (2004). Spinal cord compromise: an important but underdiagnosed condition in people with mental retardation. *Public Health Rep, 119*(4), 396-400. doi: 10.1016/j.phr.2004.05.003	X	X	Utilised charts to identify BMD from previous tests – Did not carry out BMD assessment in the study	Did not meet criteria – BMD not measured				
12	da Silva, V. Z., de Franca Barros, J., de Azevedo, M., de Godoy, J. R., Arena, R., & Cipriano, G., Jr. (2010). Bone mineral density and respiratory muscle strength in male individuals with mental retardation (with and without Down Syndrome). *Res Dev Disabil, 31*(6), 1585-1589. doi: 10.1016/j.ridd.2010.05.003	✓	✓	men only Age 20 – 35 N=45 – divided into 3 – 15 with DS; 15 ID; 15 Control group DXA – BMD		✓	9/12 Good	10/12 Good	😊
13	De Giuseppe, R., Forti, S., De Liso, F., De Vita, C., Novembrino, C., Maiavacca, R., . . . Cortelezzi, A. (2014). Evaluation of bone mineral density in down syndrome population. *Clinical Nutrition, 33*, S235.	X	X	Conference published article – no published paper	Did not meet criteria – conference paper not published				
14	Felix, M., McCubbin, J., & Shaw, J. (1998). Bone mineral	✓	✓	Premenopausal women		✓	8/12	8/12	😐

	density, body composition, and muscle strength in premenopausal women with mental retardation [corrected] [published erratum appears in ADAPT PHYS ACT Q 1999 Apr; 16(2): 201]. *Adapted Physical Activity Quarterly, 15*(4), 345-356.			Average age 29 N=32 – 16 women with ID (2 DS); 16 !! 'normal intelligence' Those with ID – Mild/mod; Independent, co-operative and ambulatory only BMD – DXA Hologic- site femoral neck			Fair	Fair	
15	Foster, B. L., Walkley, J. W., & Temple, V. A. (2001). Bone mineral density status of women with intellectual disability. *Adapted Physical Activity Quarterly, 18*(1), 49-59.	✓	✓	Women only Comparison Age avg-29 women with ID n=35 difficult to establish total N		✓	8/12 Fair	8/12 Fair	😐
16	Fowler, T. W., Akel, N. S., Schilden, J. V., Skinner, R. A., Hogue, W. R., Swain, F. L., . . . Suva, L. J. (2011). Low bone turnover may explain low bone mass in down syndrome. *Endocrine Reviews, 32*(3).	X	X	Animal study	Did not meet criteria – study on animals				
17	Gavris-Stef, M., Ferry, B., Serbanoiu, S., Hantiu, I., Lucaciu, G., Ţifrea, C., & Courteix, D. (2014). Bone Health of Subjects with Down Syndrome: Is there a Link with the Stabilometric Characteristics Measured as a Surrogate for Body Tonicity? *Procedia-Social and Behavioral Sciences, 117*, 665-671.	?	X	Dual site study DS N=193 Age range 8-37 – divided into 4 groups Compared to health non ID N=246 (n=70 had DXA) Age 10 – 61 QUS Achilles – BMD – BUA results reported but you are able to make out the results from the different groups but its very similar to the next study that is published in a different journal???	Did not meet criteria – included children				
18	Gavris, M., Courteix, D., Serbanoiu, S., Hantiu, I., Tifrea, C., & Ferry, B. (2014). Bone Tissue in Down Syndrome Patients Deteriorates Following Aging: A Study Based on Bone Ultrasound Analysis. *Journal of Medical Ultrasound, 22*(1), 29-36.	?	X	Down syndrome only Age 8-37 years N=193 – n=104 males; n=89 females Compared to N=246 Age 10 – 33 Divided into 4 groups (group 3 age 20 +/- 27months; group 4 age 33 years +/- 84months) groups 1 and 2 children up to 16 years – NO 17 – 19 years olds mentioned Difficult to establish measures of individual groups BMD - QUS	Did not meet criteria – included children, did divide out groups but unable to establish measure for groups without children				
19	Geijer, J. R., Stanish, H. I., Draheim, C. C., & Dengel, D.	✓	✓	Comparative study rated by gender and age		✓	10/12	8/12	😊

	R. (2014). Bone mineral density in adults with Down syndrome, intellectual disability, and nondisabled adults. *Am J Intellect Dev Disabil, 119*(2), 107-114. doi: 10.1352/1944-7558-119.2.107			DS v ID v non-ID Average age 43 years PWID had to be mobile BMD – DXA Total BMD given and compared with groups			Good	Good	
20	Glick, N. R., Fischer, M. H., Heisey, D. M., Leverson, G. E. & Mann, D. C. (2005) Epidemiology of fractures in people with severe and profound developmental disabilities. *Osteoporos Int,* **16,** 389-96.	X	X	Did not report BMD Fracture rates only Chart review	Did not meet criteria – chart review only did not measure BMD				
21	Gonzalez-Aguero, A., Vicente-Rodriguez, G., Gomez-Cabello, A., & Casajus, J. A. (2013). Cortical and trabecular bone at the radius and tibia in male and female adolescents with Down syndrome: a peripheral quantitative computed tomography (pQCT) study. *Osteoporos Int, 24*(3), 1035-1044. doi: 10.1007/s00198-012-2041-7.	X	X	Comparative DS v healthy subjects BMD – pQCT Age range 11 – 20 years Mean age 15 years - rejected on age	Did not meet criteria - children				
22	Guijarro, M., Valero, C., Paule, B., Gonzalez-Macias, J., & Riancho, J. A. (2008). Bone mass in young adults with Down syndrome. *Journal of Intellectual Disability Research, 52*(3), 182-189.	✓	✓	comparative study PWID – age range 18 – 45 n-39 ; 18 males , 21 females Non-ID age range 19 – 78 n=78 BMD – DXA Hologic Vol BMD calculated by formulae Definition of ID and DS noted Had to be mobile		✓	7/12 Fair	7/12 Fair	😐
23	Jaffe, J. S., Timell, A. M., Elolia, R., & Thatcher, S. S. (2005). Risk factors for low bone mineral density in individuals residing in a facility for the people with intellectual disability. *Journal of Intellectual Disability Research, 49*(Part 6), 457-462.	?	X	N=211 men-132 women 79 all levels of ID BMD Sahara Hologic – QUI calculated by formulae to compare with DXA 77.2% osteoporotic or osteopenic	This study was a combination of the next 2 articles by the same author with the same data utilised				
24	Jaffe, J. S., & Timell, A. M. (2003). Prevalence of low bone density in institutionalized men with developmental disabilities. *Journal Of Clinical Densitometry: The Official Journal Of The International Society For Clinical Densitometry, 6*(2), 143-147.	?	✓	men only N=135 Mean age 51.9 years QUS Sahara Hologic – QUI calculated from BUA and SOS compared to reference population men age 20 – 89 years from another study Seems to be from published study above	✓		9/14 Fair	9/14 Fair	😐
25	Jaffe, J. S., Timell, A. M., & Gulanski, B. I. (2001). Prevalence of low bone density in women with developmental disabilities. *Journal Of Clinical Densitometry: The Official Journal Of The International Society For Clinical*	?	✓	Women only Average age >60 years N=80 QUS Sahara Hologic	✓		FF	FF	😐

	Densitometry, 4(1), 25-29.			Almost identical article as previous – same study					
26	Jasien, J., Daimon, C. M., Maudsley, S., Shapiro, B. K., & Martin, B. (2012). Aging and Bone Health in Individuals with Developmental Disabilities. *International Journal of Endocrinology*. doi: 10.1155/2012/469235	X	X	Chart review good summary of articles and other studies of interest	Did not meet criteria – chart review				
27	Leslie, W. D., Pahlavan, P. S., Roe, E. B., & Dittberner, K. (2009). Bone density and fragility fractures in patients with developmental disabilities. *Osteoporosis International, 20*(3), 379-383. doi: 10.1007/s00198-008-0678-z	✓	✓	N=107 PWID n=84 had measurement with in osteoporotic range (78.5%) Age >20 years Peripheral DXA - PIXI GE Lunar of forearm and heel Data given in mean	✓		9/14 Fair	9/14 Fair	😐
28	Lin, L.-P., Hsu, S.-W., Yao, C.-H., Lai, W.-J., Hsu, P.-J., Wu, J.-L., . . . Lin, J.-D. (2015). Risk for osteopenia and osteoporosis in institution-dwelling individuals with intellectual and/or developmental disabilities. *Research in developmental disabilities, 36*, 108-113. doi: 10.1016/j.ridd.2014.09.022	✓	✓	N=188 PWID Men n=115; Females n=69 age range 18 – 72 years QUS – DMS Pegasus smart bone densitometer	✓		9/14 Fair	9/14 Fair	😐
29	Lohiya, G., Tan-Figueroa, L., & Iannucci, A. (2004). Identification of low bone mass in a developmental center: finger bone mineral density measurement in 562 residents. *Journal of the American Medical Directors Association, 5*(6), 371-376.	X	X	Focus on # No BMD measured	Did not meet criteria – chart review				
30	May, P. (2015). Prevention, Evaluation, and Management of Low Bone Mineral Density (BMD) Encountered in Adults with Developmental and Intellectual Disabilities. *Exceptional Parent, 45*(2), 38-40.	X	X	Chart review No BMD measurements undertaken	Did not meet criteria – chart review				
31	McKelvey, K. D., Fowler, T. W., Akel, N. S., Kelsay, J. A., Gaddy, D., Wenger, G. R., & Suva, L. J. (2013). Low bone turnover and low bone density in a cohort of adults with Down syndrome. *Osteoporos Int, 24*(4), 1333-1338. doi: 10.1007/s00198-012-2109-4	✓	✓	DS only compared to 8 healthy normal volunteers N=30 males and females Males 17; Females 13 Separates out those with DS Age 19 – 52 years DXA Hologic Discovery A Bone densitometer - BMD		✓	4/14 Poor	4/14 Poor	😐
32	Mergler, S., Bastiaanse, L. P., Echteld, M. A., & Evenhuis, H. M. (2012). Prevalence and associated factors of osteoporosis in older adults with intellectual disabilities. *Journal of Intellectual Disability Research, 56*(7-8), 757.	X	X	Conference published article – no published paper	Did not meet criteria – conference paper not published				

33	Mergler, S., Bastiaanse, L. P., Wielders, J. P. M., Kingma-Thijssen, J. L., Echteld, M. A., & Evenhuis, H. M. (2012). Vitamin D levels in an ageing ID population. *Journal of Intellectual Disability Research, 56*(7-8), 754.	X	X	Conference published article – no published paper	Did not meet criteria – conference paper not published				
34	Milberger, S., LeRoy, B., Lachance, L., & Edelson, G. (2002). Osteoporosis and women with Down syndrome. *Journal of Intellectual and Developmental Disability, 27*(4), 273-280. doi: 10.1080/1366825021000055808	✓	✓	N=23 Female only DS only DXA – BMD Expresses results at sites – spine, hip and proximal forearm	✓		7/14 Fair	7/12 Fair	😐
35	Múgica, I., Ansa, J., Sistiaga, F., Zabalza, R., Zubillaga, P., Merino, A., Vidal, C. (2002). [Digital computerized absorptiometry in the diagnosis of osteoporosis in a group of the severely mentally retarded]. *Nutrición Hospitalaria, 17*(4), 213-218.	X	X	Abstract was in English but article is in Spanish	Did not meet criteria – foreign language paper				
36	Petrone, L. R. (2012). Osteoporosis in adults with intellectual disabilities. *Southern Medical Journal, 105*(2), 87-92. doi:.1097/SMJ.0b013e3182427042	X	X	Chart review only no measurement	Did not meet criteria – Chart review				
37	Ray, J. G., Papaioannou, A., Ioannidis, G., & Adachi, J. D. (2002). Anticonvulsant drug use and low bone mass in adults with neurodevelopmental disorders. *QJM: An International Journal of Medicine, 95*(4), 219-223. doi: 10.1093/qjmed/95.4.219	✓	✓	N=273 Low BMD 22.7% QUS Sahara Clinical Bone Sonometer - Hologic	✓		4/14 Poor	4/14 Poor	😐
38	Robinson, L., Zarcone, J., Fortuna, R., Nobis, K., Meccarello, J., & Davidson, P. W. (2012). Health outcomes in adults with Praeder-Willi syndrome. *Journal of Intellectual Disability Research, 56*(7-8), 658.	X	X	Conference published article – no published paper	Did not meet criteria – conference paper not published				
39	Roselló, L., Torres, R., Boronat, T., Llobet, R., & Puerto, E. (2004). Osteoporosis prevalence in a Down syndrome population, measuring different parameters. *SD Revista Medica Internacional sobre el Sindrome de Down, 8*(2), 18-22.	✓	✓	N=51 All employed in sheltered workshop BMD – DXA Lunar dual –photon densitometer 45.1% osteoporosis; 29.4% osteopenia	✓		8/14 Fair	7/12 Fair	😐
40	Ryder, K. M., Williams, J., Womack, C., Nayak, N. G., Nasef, S., Bush, A., Carbone, L. (2003). Appendicular fractures: a significant problem among institutionalized adults with developmental disabilities. *American Journal Of Mental Retardation: AJMR, 108*(5), 340-346.	✓	✓	Comparative study matching 23 PWID with history of fracture with 23 PWID no history of fracture QUS –Sahara Hologic n=13 who refused to have QUD Compared n=16 # with n=17 non-# (#=fractured)		✓	8/12 Fair	8/12 Fair	😐

41	Schmidt, E. V., Byars, J. R., Flamuth, D. H., Schott, J. J., & Sever, C. M. (2004). Prevalence of low bone-mineral density among mentally retarded and developmentally disabled residents in intermediate care. *Consultant Pharmacist, 19*(1), 45-51.	✓	✓	Starts off stating N=568, states N=360 for BMD testing –excluded n=119 according to their exclusion criteria foot deformity etc = final N=241; n=157 females (67%) BMD – PIXI Age 20 – 91 Average age 45 years Osteoporosis 65.1%; Osteopenia 13.3%	✓		9/14 Fair	9/14 Fair	😐
42	Sepúlveda, D., Allison, D. B., & Gomez, J. E. (1995). Low spinal and pelvic bone mineral density among individuals with Down syndrome. *American Journal on Mental Retardation, 100*(2), 109-114.	✓	✓	DS only 11 males and 4 females N=15 compared to 25 control subjects without DS DXA – BMD – Lunar DPX scanner at 3 sites arm, pelvis and spine Results given in mean gms/cm2 difficult to establish but all lower / poorer than controls		✓	6/12 Fair	7/12 Fair	😐
43	Snoeijen-Schouwenaars, F., Tan, I., & Verschuure, P. (2014). Feasibility of whole body vibration training in people with epilepsy, osteopenia and an intellectual disability. *Epilepsia, 55*, 201.	X	X	Conference published article – no published paper	Did not meet criteria – conference paper not published				
44	Srikanth, R., Cassidy, G., Joiner, C., & Teeluckdharry, S. (2011). Osteoporosis in people with intellectual disabilities: a review and a brief study of risk factors for osteoporosis in a community sample of people with intellectual disabilities. *J Intellect Disabil Res, 55*(1), 53-62. doi: 10.1111/j.1365-2788.2010.01346.x	X	X	Retrospective chart review – focus on risk factors No BMD measures	Did not meet criteria – Chart review				
45	Sustrova, M., Krivosikova, Z., Spustova, V., & Stefikova, K. (2009). Is early onset of osteoporosis in women and men with Down syndrome based on different factors? *Bone, 44*, S412.	X	X	Conference published article – no published paper	Did not meet criteria – conference paper not published				
46	Tyler, C. V., Jr., Snyder, C. W., & Zyzanski, S. (2001). Screening for osteoporosis in community-dwelling adults with mental retardation-errata. *Mental Retardation, 39*(1), 39-39. doi: 10.1352/0047-6765(2001)039<0039:E>2.0.CO;2	✓	✓	N=107 age 40 – 60 mean age 47 years BMD – Lunar PIXI - QUS of calcaneus 21% osteoporotic 34% osteopenic	✓		10/14 Good	10/14 Good	😊
47	Vice, M. A., Nahar, V. K., Bass, M., & Ford-Wade, A. (2012). Risk Factors for Low Bone Mineral Density in Institutionalized Individuals with Developmental Disabilities. *Medicine and Science in Sports and Exercise, 44*,	X	X	Chart review Very robust data collection method – strong article	Did not meet criteria – chart review				

	707-708.								
48	Wagemans, A., Fiolet, J., Van Der Linden, E., & Menheere, P. (1998). Osteoporosis and intellectual disability: is there any relation? *Journal of Intellectual Disability Research, 42*(5), 370-374.	?	✓	Calls the participants bed ridden with ID BMD – DXA 75% osteoporosis Very scant article –bare minimum information	✓		3/14 Poor	4/14 Poor	
49	Wallen, E. F., Müllersdorf, M., Christensson, K., Malm, G., Ekblom, Ö., & Marcus, C. (2009). High prevalence of cardio-metabolic risk factors among adolescents with intellectual disability. *Acta Paediatrica, 98*(5), 853-859.	X	X	Adolescents N=156 Mean age 18.6years All school going	Did not meet criteria – mostly children				
50	Zylstra, R. G., Porter, L. L., Shapiro, J. L., & Prater, C. D. (2008). Prevalence of osteoporosis in community-dwelling individuals with intellectual and/or developmental disabilities. *Journal of the American Medical Directors Association, 9*(2), 109-113.	✓	✓	N=298 community dwelling PWID Age range 6 – 90 years (however on table 1 there is only one participant under the age of 19 years) Scans appear to have been taken over many years Jan 98 – Jan 06. N=298 scans – DXA Does divide out results in age groups and you can pull-out results those over 18 years Osteoporosis 17.1%	✓		8/14 Fair	11/14 Good	DNGF DXA results appear to be from charts

Quality Assessment Criteria -

Rating	Case-Control Studies	Observational Cohort & Cross-Sectional Studies	Action
	Confirmed Yes	Confirmed Yes	
Good	9-12	10-14	Move to data extraction
Fair	6-8	7-9	Adjudicate with 3rd independent assessor
Poor	≤5	≤6	Reject
Other	CD, NR, NA*	CD, NR, NA*	Consider adjudication

Filter results following first review

Going forward for Quality Assessment (QA) – 22
Query first before forwarding to QA – 9
Rejected - 19

Filter results following second review

Going forward for Quality Assessment – 27
Rejected – 21
Agreed: EB_____✓_____ MOD_____✓_____

QA
6 articles selected – see results above

Appendix 3:
Data Extraction Form

Data extraction tool for the '*Prevalence of Osteoporosis among adults with an intellectual disability*'

Number	Author & Year	Study design	Age	Population								Specific living circumstance				Type of equipment utilised to establish prevalence	Prevalence data	Comments & controls
		Case control [CC] Observational [OC]		Men only	Women only	Post-menopausal Women	Men and women	Level of ID	aetiology specific	Down syndrome[DS]	Other aetiology ID	Independent	Community group home	Residential or congregated	None specified	DXA QUS	% or g/cm2 (raw data)	
1	Angelopoulou et al 1999	CC	♂ age 26.22 ±4.45 ♀23.65 ±3.23				✓	Not specified		9♂ 13♀		✓				DXA – Lunar DPX v3.6	BMD 0.978g/cm² ♂ t-score; mean -2.62 ± 0.82 - 25% ♀ t-score: - 1.56±1.02 14%	27 healthy controls
2	Angelopoulou et al 2000	CC	[DS] ♂age 23.88 ±4.22 ♂23.5 ±3.59	✓				mod/mild		8	8	✓				DXA – Lunar DPX	BMD 0.926g/cm² t-score mean_-2.66 ±0.07 [DS] BMD [ID group] 1.228 g/cm² ±0.14 t-score mean - 0.15 ±0.52	10 controls ♂age 25.0 ±3.3 t-score mean-0.06 ±0.29
8	Center et al 1998	OC	♀ age 35±7 ♂age 35±9				✓	All		[21% of total pop. - DS]	53♀ 41♂		✓ 90%	✓		DXA – Norland XR-26	BMD mean 0.97 g/cm² ±0.19 ♀ 1.01 g/cm² ±0.17 ♂	

12	da Silva et al 2010	CC	[DS] ♂age 28.75 ±7.71 [ID] ♂28.14 ±5.61	✓				mod/mild		15	15			✓		DXA – DPX-IQ v4.6A	[DS] BMD mean 0.94 g/cm² ±0.11 [ID] BMD 1.26 g/cm² ±0.14	15 ♂ controls (non-ID) age 24.57 ±3.87 no differences between BMD of ID and control group
19	Geijer et al 2014	CC	[DS] age 43.7 ±5.5				✓			33	33					DXA- does not specify (DNS)	[DS] BMD mean 1.10 g/cm³ ± 0.11 [ID] BMD 1.20 g/cm³ ±0.14	[all PWID were independently ambulant] 33 controls (non-ID) age 43.7 ±5.9 BMD 1.28 g/cm³ ±0.14
46	Tyler et al 2000	OC	age 47 ±5.6				✓	Not specified			46♀ 61♂		✓			DXA – Calcaneus Lunar PIXI	21% osteoporosis 34% osteopenia 45% Normal BMD 0.48 g/cm² ±0.14 0.42 g/cm² ±0.12	

Appendix 4:
IDS-TILDA Wave 1 Invitation Pack

a. Cover letter
b. Letter of support from service providers
c. Family support pack
d. Accessible consent form

Intellectual Disability Supplement to TILDA
The University of Dublin, Trinity College
School of Nursing & Midwifery
24 D'Olier Street, Dublin 2.
Tel: + 353 1 896 3186/3187
Fax: + 353 1 896 3001 Email: idstotilda@tcd.ie

An Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing

<Insert date>



Dear Participant,



We are writing to tell you about a large study we will be carrying out on growing older with intellectual disability in Ireland.



This is a very important study which will involve 800 people with intellectual disability.



It will help us to learn more about what is important to people as they grow older.



We will learn about people's health, work and lifestyles.



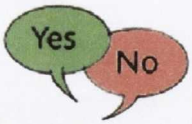
We would like to invite you to take part in this study.
We are sending you an information booklet about this study and a consent form.

A consent form is something a person signs which means that they are happy to help and take part.

Please read the information booklet or ask someone to help



you to read it. This will help you to understand the study better.



It is your choice to take part in the study.



If you would like to take part, please read and sign the consent form, or ask someone to help you.



We would be delighted if you decided to take part in this study. If you want to ask us any questions then please cal us on (01) 8963186 or 8963187.

We look forward to hearing from you.



Yours sincerely,

Prof. Mary McCarron.

Professor Mary McCarron
Principal Investigator
Intellectual Disability Supplement to TILDA

4b – Service Provider Support Letter

<Organisation Logo/Headed Paper>

Sample Letter

Dear Participant/Family Member,

Please find attached information regarding a new national study recently launched by Professor Mary McCarron at Trinity College Dublin on Ageing in Persons with an Intellectual Disability in Ireland.

<Name of Organisation> very much supports this national study and feels it represents a unique opportunity to explore the health, social and environmental circumstances of people with intellectual disability as they age, and allow comparisons to be made with the general population.

You/your family member are being invited to participate in this study. The decision about whether to do so or not is entirely your own. Whatever decision you make will in no way affect the current or future services or supports offered to you/your family member.

The study is voluntary, but it is important that all of the people who are selected take part, if at all possible, so that the results give an accurate picture of people with intellectual disabilities lives as they age.

Yours sincerely,

<Signature>

Chief Executive Officer

Intellectual Disability Supplement to TILDA
The University of Dublin, Trinity College
School of Nursing & Midwifery
24 D'Olier Street, Dublin 2.
Tel: + 353 1 896 3186/3187
Fax: + 353 1 896 3001 Email: idstotilda@tcd.ie

An Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing

<Insert date>

Dear family member/guardian,

The Intellectual Disability Supplement to TILDA is the most comprehensive study on ageing in persons with intellectual disability ever to be undertaken in Ireland. For the first time, there is a large and growing population of older people with intellectual disability in Ireland and elsewhere. Although this is an incredible success, we know little about the ageing of this population group.

This study will explore the health, lifestyles, living circumstances and quality of life for about 800 people with intellectual disability as they grow older. It will also see how their circumstances change over a 10-year period. The study is being carried out by Trinity College Dublin and is part of a major Irish longitudinal study on ageing (TILDA) in the general population, which is led by Professor Rose Anne Kenny.

We have chosen your family member/person with intellectual disability to take part in this study. This is voluntary, however it is a very important study and we will get better information if we have a large number of people taking part.

Please find attached an information leaflet about the study and what it involves. If your family member/person with intellectual disability decides to take part in this study, an interviewer with a lot of experience of working with people with intellectual disability will arrange a suitable time to conduct the interview with them. We hope you agree to your family member/person with intellectual disability taking part in this study. If so, please sign the attached agreement form and return it in the stamped addressed envelope enclosed. If you have any questions, please do not hesitate to contact me or a member of the research team on 01-896 3186/3187.

Yours sincerely,

Prof. Mary McCarron.

Professor Mary McCarron
Principal Investigator
Intellectual Disability Supplement to TILDA

What is the study about?

The Intellectual Disability Supplement to the Irish Longitudinal Study on Ageing (IDS to TILDA) is the most important study on ageing ever undertaken in Ireland.

It will look at the health, lifestyles and quality of life of about 800 people with intellectual disability as they grow older. It will also observe how their circumstances change over a 10-year period.

The information will be used to develop suitable health and social services that will benefit all people with intellectual disability as they age in Ireland.

We are inviting people with intellectual disability to take part in this study which we are carrying out.

How are participants selected for the study?

People who are registered on the National Intellectual Disability Database (NIDD) and are aged over 40 years may be selected to take part in the study. The sample will be randomly selected from this database.

Does the person with intellectual disability that is selected have to take part?

No. Taking part is voluntary. However, this is a very important study and the information we collect will be better if we have a large number of people taking part. We are inviting men and women aged 40 years and older with an intellectual disability to take part. The person with intellectual disability selected can decide to withdraw from the study at any time.

What if the person with intellectual disability does not take part?

If the person with intellectual disability does not take part or later decides to withdraw, then their decision will in no way affect the current or future services or supports offered to them.

What does the study involve?

A researcher with experience of working with people with intellectual disability will visit the person with intellectual disability and ask questions about their health, lifestyle, living circumstances and quality of life.

We understand that some people may not be able to give this information on their own and may need support. If this is the case then the person with intellectual disability may ask someone (for example, a family member, guardian or key worker) to support them in giving this information.

We will also ask the person with intellectual disability to try and carry out some tasks such as:

- brushing their hair
- writing their name
- naming some colours

The interview will take about 90 minutes. However, this will be different for each person.

At the end of the interview we will arrange a suitable time to take some important physical measurements such as height and weight. We will use these results to find out much more about the health of people with intellectual disability than we could by just asking questions about diet and lifestyles alone. The measurements will take about 15 minutes.

Are there any risks involved?

There are no known risks involved in this study. We will explain each stage of the study to the person with intellectual disability using suitable information booklets and show cards.

At all times, the well-being of the person with intellectual disability will take priority over the research activities. If the person with intellectual disability tells us or indicates that they do not want to take part, or if they decide to withdraw, we will fully respect their decision.

Is the study confidential?

Yes. We will treat all the information from the study as strictly confidential and we will not publish the name of the person with intellectual disability or pass it on to anyone outside of the research team. The Faculty of Health Sciences Research Ethics Committee, of Trinity College, Dublin, has approved this study after ensuring that proper safeguards are in place.

Who is involved in the study?

This study has been set up by people who have a lot of experience of working with people with intellectual disability.

The advisory team consists of:

- Family Members
- Researchers
- Nurses
- Physicians
- Psychologists
- Psychiatrists
- Service Providers

People with intellectual disability also play an important role in our advisory team through participation in consultative workshops. These workshops will be ongoing with groups throughout the country as the study progresses.

Where can I get more information?

If you have any questions, or if you want more information about the study, please do not hesitate to call or email one of the contacts listed below.

Professor Mary McCarron
Principal Investigator
Phone: 01- 896 3186/3187
Email: mccarrm@tcd.ie

Ms Janet Swinburne
Project Manager / PhD Student
Phone: 01- 896 3186/3187
Email: swinburj@tcd.ie

Or write to:

Intellectual Disability Supplement to TILDA,
The University of Dublin,
Trinity College,
School of Nursing & Midwifery,
24 D'Olier Street,
Dublin 2.

Fax: 01- 8963001
Email: idstotilda@tcd.ie

The Intellectual Disability Supplement to TILDA is funded by the Health Research Board.

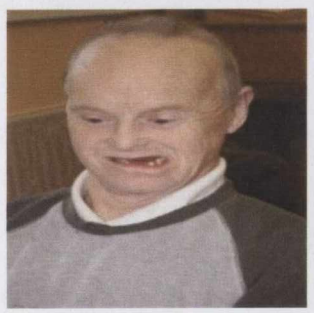
HRB Health Research Board

tilda The Irish Longitudinal Study on Ageing

An Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing

An Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing (IDS to TILDA)






Trinity College Dublin

Consent form

Please read the information below and sign this consent form if you wish to take part in this study.

I agree with the following statements:

Please tick



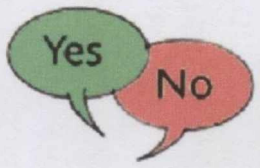
I have gone through the information about this study ☑



I knew who to contact if I had any questions. ☐



Any questions that I might have had were answered. ☐



I know that it is my choice to take part in this study. ☐



I understand that I will be asked questions about my: ☐

- life
- health
- work
- friends

and things I like to do.

Please tick



I understand that I do not have to answer questions I don't feel happy with. ☐



As part of the study, I know that I will be asked to try and do some things such as write my name. ☐



I understand that I will be asked to have some measurements taken such as my height and weight. ☐



I understand that I can stop taking part in this study when I want to. I do not have to give a reason. ☐



I understand that all information I give during this study will be kept safe and private. I will not be named in any reports. ☐



I understand that there are no known risks with this study. ☐



I am happy to take part in this study. ☐

Your consent

Your name: _____ Your phone number: _____

Your address:

_____

_____

_____

_____

_____

Please tick ☑

When is the best time to contact you?

Morning time ☐

Afternoon time ☐

Evening time ☐

Please sign your name: _____ Date: _____

The person supporting you

I have supported the person named above to fill out this form. I believe they understand the information and have freely agreed to take part in this study.

Print name: _____

Relationship to the person named above: _____

Phone number:_____

Signature: _____ Date:_____

Please return this consent form in the envelope provided.

Intellectual Disability Supplement to TILDA, The University of Dublin, Trinity College, School of Nursing & Midwifery, 24 D'Olier Street, Dublin 2
Tel: +353 1 8963186/8963187 Fax: +353 1 8693001 Email: idstotilda@tcd.ie

Appendix 5:

Keeping in touch strategy

Keeping in Touch Strategy - The Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing (IDS-TILDA)

It is imperative for longitudinal studies that a solid 'keeping in touch' strategy is employed to maintain the link with the participant community. Maintaining connectivity with participants ensures they remain central to the study, are acknowledged for their contribution and preserve study numbers. Along with hybrid publications, IDS-TILDA has created such platforms for their study participants to ensure a robust 'Keeping in touch' strategy.

WEBSITE

- Online platform that provides up to date news on IDS-TILDA activity, including reports, accessible videos and strategy actions
- Website is updated monthly

NEWSLETTERS

- Bi-annual newsletter published with up to date IDS-TILDA activity
- Each individual participant receives a newsletter
- IDS-TILDA holds competitions and events advertised through the website and newsletter for all participants

PERSONAL CONTACT

- Each participant receives a personalised Easter and Christmas card every year from the PI and study team
- Each participant receives a phone call from the study team prior to recommencement in the field
- This contact is ongoing and ensures a link with participants, and confirmation of their address

ROADSHOW

- Ensuring participants, families and support staff realise the benefit of the research IDS-TILDA run seminars and events in services
- Such events are ongoing and it is anticipated that this activity will increase as the next study wave nears. These events are imperative in disseminating results, maintaining the sample and demonstrating appreciation.

***Source**: The Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing.*

Appendix 6:

Wave 2 Consent form

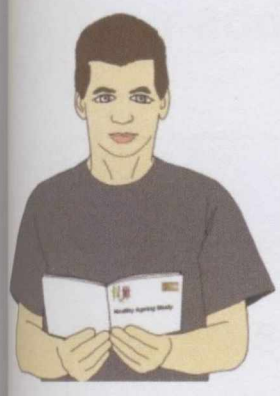
Wave 2 Information Booklet

CONSENT FORM

Please read the information below and sign this consent form if you wish to take part in this second wave of the study.

I agree with the following statement

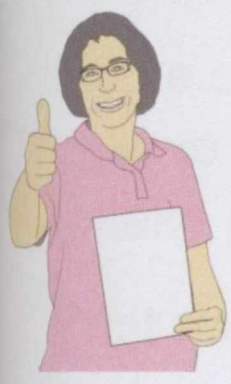
Please tick ✓



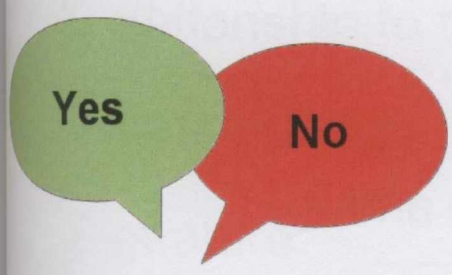
I have gone through the information about this study ☐



I know who to contact if I have any other questions. ☐



Any questions that I might have had were answered. ☐



I know that it is my choice to take part in this study. ☐

I understand this study is for ten years and I will be visited again by a researcher from Trinity College Dublin. ☐

Trinity College Dublin



I understand that I will be asked questions about my:

- life
- health
- work
- friends

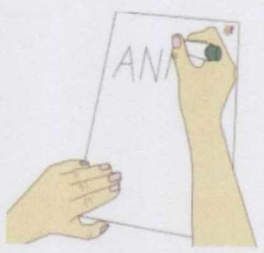
and things I like to do.



I have gone through the information about this study



I understand that I do not have to answer questions I don't feel happy with.



As part of the study, I know that I will be asked to try and do some things such as write my name.



I do understand that I can stop taking part in this study when I want to.

I do not have to give a reason.



I understand that all information I give during this study will be kept safe and private.

I will not be named in any reports.



I understand that there are no known risks with this study.

YOUR CONSENT

Your name: _____

Your phone number: _____

Your address: _____

_____

_____

Please sign your name:

_____

Date: _____

THE PERSON SUPPORTING YOU

I have supported the person named above to fill out this form. I believe they understand the information and have freely agreed to take part in this study.

Print name: _____

Relationship to the person named above: _____

Phone number: _____

Signature: _____

Date: _____

Please return this consent form to the field researcher before the interview commences.

IDS-TILDA, The University of Dublin, Trinity College, School of Nursing & Midwifery, 24 D'Olier Street, Dublin 2

Tel: +353 1 8963186/8963187 Fax: +353 1 8693001 Email: idstilda@tcd.ie

TRINITY COLLEGE DUBLIN
COLÁISTE NA TRÍONÓIDE
THE UNIVERSITY OF DUBLIN

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An Intellectual Disability Supplement to
The Irish Longitudinal Study on Ageing



Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing (IDS-TILDA)

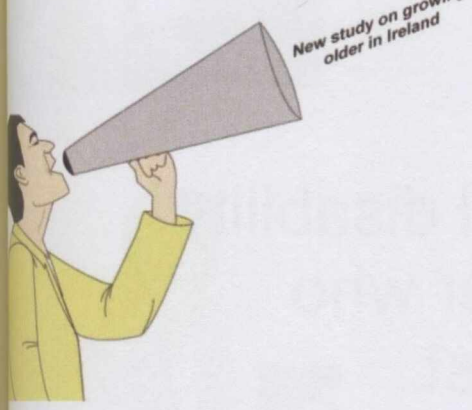
WAVE 2
INFORMATION BOOKLET

This study is being carried out by Trinity College Dublin

ear Participant,

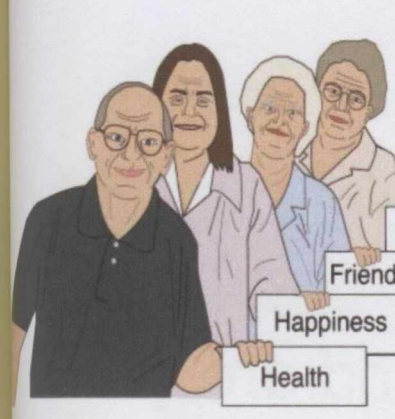
/e are delighted to tell you we will be starting the
terviewing again from April. This booklet reminds you about
)S-TILDA and tells you about some of the new things we
ould like to ask when we visit.

. What is this study about?



This study is about growing older with an intellectual disability in Ireland.

Trinity College Dublin is doing this study.



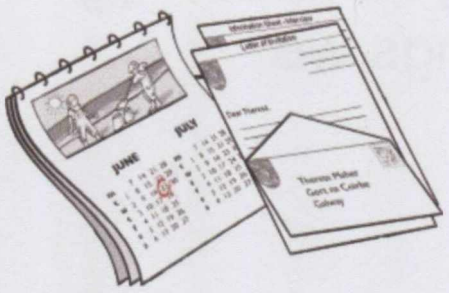
This study will help us to understand what is important to people



It will help us to learn about the

- health
- well-being
- and lifestyles

of people as they get older.



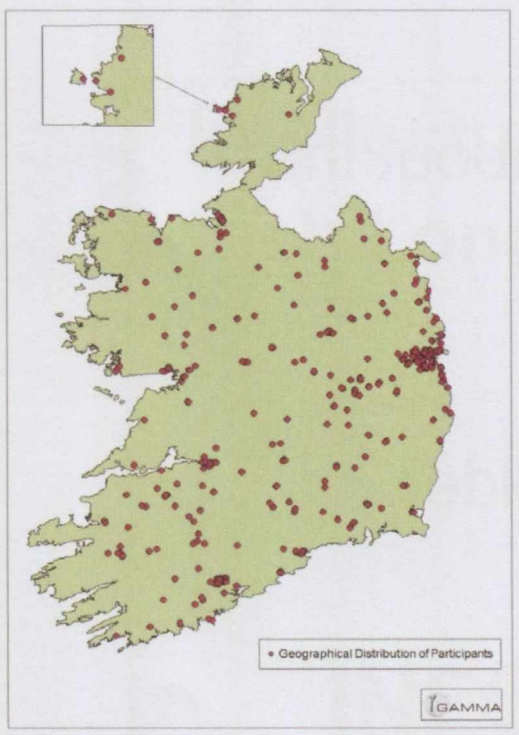
This is your second visit. We will see what has changed for you since your first interview.

2. Who is taking part in the study?



People with intellectual disability aged 40 years and over who were chosen for the first interview.

3. Who is taking part?



People from all over Ireland are taking part in the study.

You were one of the people selected to take part.

Thank you for answering the questions when we last visited.

What will I be asked this time?

You will be asked some of the same questions we asked before and some new questions about



- Activities you do everyday



- Your health and well-being



- Your friends



- Your family



- Interests and hobbies

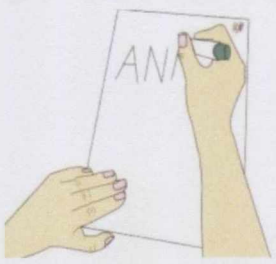


- Work and retirement



- Your home

You can choose someone to help you answer questions



You will be asked to do some things like write your name



The researcher will put your answers into a laptop computer at the interview.

We will also be doing a health assessment.



This involves taking measurements, for example your height, your weight and your blood pressure.



We will also measure your bone strength and your grip strength.





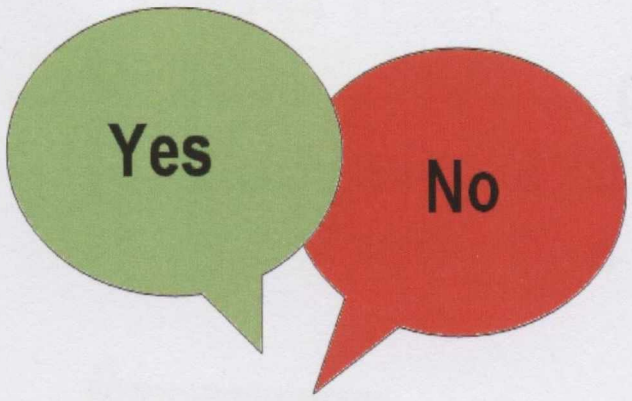
A researcher will be in touch with you to tell you more about the health assessment.

Who will visit you?

Trinity College Dublin

Researchers working on the study from Trinity College Dublin will visit you.

6. Do you have to take?



No. It is your choice to take part in the study. You can also change your mind and stop taking part at any time.

Your decision will not affect the support you receive now or in the future.

7. How long will it take?



The interview will take about 90 minutes.

The Health Assessment will take about 30 minutes.



If the interview is too long you can stop and take a break

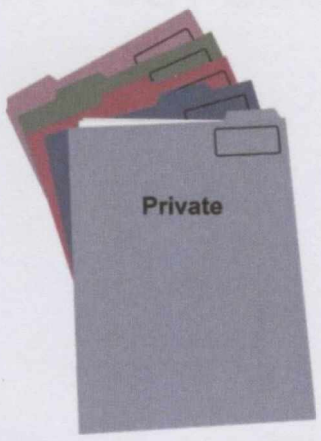
If you want we can call back another day

Are there any risks involved?



We do not know of any risks to you being involved in this study.

Will anyone else know what you tell us?



No. We will keep all your personal information private. Your name will not be in any report.

What should you do next?



A researcher will be in contact with you soon to see when it suits for us to come and visit you.

Is there anything else you need to think about?

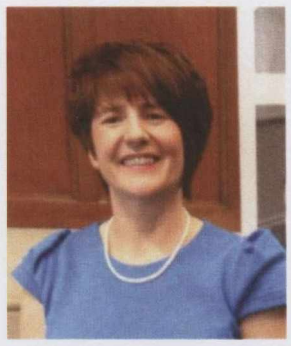


Yes. You need to think about:
Where and when we can meet for the interview.
If you want to bring someone with you to the interview.

12. Who do you contact if you have any questions?

?

If you need more information or if you have any questions, please contact:



Professor Mary McCarron
Principal Investigator

Phone: 01-8963187 or 01-8963186

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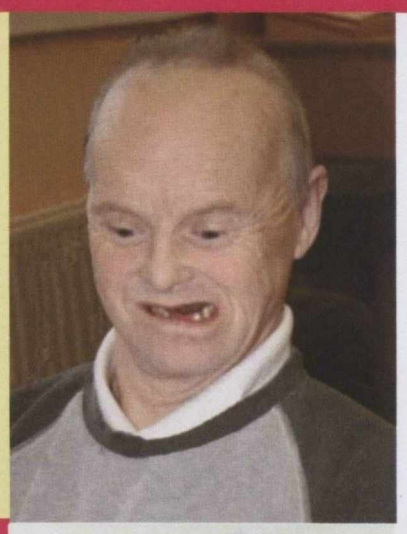
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e Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing is
nded by the Health Research Board and the Department of Health and Children

The IDS-TILDA Health Fair





Healthy Ageing Happy

An Intellectual Disability Supplement to
The Irish Longitudinal Study on Ageing





CALLING ALL IDS-TILDA PARTICIPANTS

Over the next few months we will ask you to take part in the Health Fair. This will mean taking some health measures such as your blood pressure.

These measures will help us understand if people's health change as they grow older. It will help us understand what people need in order to live a healthy happy life as they grow older.

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Department of Health & Children
HRB Health Research Board
tilda The Irish Longitudinal Study on Ageing

Wave 2 of the Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing has begun.

From Wave 1 we have begun to get a good idea of what it is like for people with an intellectual disability in Ireland to grow older. We have written about what we found out and made the DVD to let you all know of these important results. You have made this possible by taking part in the interviews. As part of the next stage of the study we are going to do a health fair.

What is the Health Fair about?

The Health Fair is about taking health measures like blood pressure or weight. These measures will give us a very clear picture of the health of people in the study. We will be able to build an overall picture of how people's health is like as they grow older with ID.

What is involved?

A researcher will contact you to make an appointment. The researcher will come to where you live to do the measurements. Sometimes she will do the health measures in a central place like your local service provider's. There are 7 health measures. You will be asked to have your blood pressure taken, weight, height, waist and hips measured. We will also measure your grip strength, get up and go and your bone density.

Do I have to take part?

No, you do not have to take part in the health assessment. The researcher will explain all the measurements. She will ask you for your permission. The researcher will also ask you for your permission before each measurement.

Will it take long?

The full assessment takes about 30 minutes. You will get your results on the day.

Thank you for your help.

Appendix 8

Health Fair Protocol

An Intellectual Disability Supplement to
the Irish Longitudinal Study on Ageing

INTELLECTUAL DISABILITY SUPPLEMENT TO THE IRISH LONGITUDINAL STUDY ON AGEING

The Health Fair Protocol

Objective measurements field researcher guidelines

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1. Introduction

The Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing (IDS-TILDA) is a major ageing study which will provide high quality research relating to older people with intellectual disability as they age in Ireland. IDS-TILDA will contribute to providing a comprehensive and accurate picture of the characteristics, needs and contributions of older people with ID in Ireland which will be invaluable to policy makers and public sector service planners and to services engaged in activities seeking to enhance the social and community integration of people with ID. To that end IDS-TILDA are including physical assessments in Wave 2. To avoid any negative connotations associated with medical measurement the health assessment process will be referred to as The Health Fair©. To ensure all participants have an opportunity to participate the Health Fair© will take place at a time date and venue convenient to them (to the best of the assessor's ability, facilities permitting). Due to the requirements of space and the logistics of setting up equipment the aim is to identify central locations for setting up the Health Fair© in liaison with the service provider gatekeeper.

The Health Fair© is part of the data collection of Wave 2 however will be conducted separate to the main face to face interview to reduce any potential burden. Data collection is comprised of three sections and will occur in the following manner

- The pre-interview questionnaire
- The interviewer administered interview (CAPI)
- The physical assessment

The physical assessment is new to the IDS-TILDA and therefore it is difficult to establish how the participants will respond however results from similar studies (TILDA, ELSA) suggest that most participants find the physical assessment a positive aspect to the study and are happy to take part. Considering the individual needs of the participants the assessor administering the physical assessment has specific training in the area, has extensive and considerable experience working with people with ID and is skilled in the research design of IDS-TILDA.

2. The Health Fair©

The Health Fair©, which incorporates all 8 health measurements, is an integral part of the IDS-TILDA study. The assessments include; grip strength, waist measurement, hip measurement, timed up and go (TUG), blood pressure (2 sitting and 2 standing), height, weight and quantitative heel ultrasound (QUS). The assessments will not take any longer than 30 minutes. The process for setting up the health assessment is as follows

2.1 The Health Assessment Process

1. •Inform all participants of Wave 2 commencement and the inclusion of health assessment – this will included poster advertisement and written explanation
2. •Link with gatekeeper from services, arrange site for Health Fair© convenient to participants and dates for potential appointments
3. •Contact participant and their support worker and agree appointment dates, post easy read information cover letter and easy read explanatory booklet
4. •Reconfirm appointment on day of assessments (prior to travelling), liaise with gatekeeper
5. •Arrive with minimum 30 minutes for set up, quality checks and equipment calibration
6. •At time of health assessment it is imperative that no measurements are attempted until consent has been obtained, for those who are non-verbal obtain assent and co-operation and direction of support worker.
7. •Participant co-operation is completely voluntary for each assessment, employ process consent

2.2 Underpinning Philosophy

The design and management of the Health Fair© was conducted through the key principles of Emancipatory Philosophy. This was specifically chosen as it portrays the core values required to foster the capacity of each participant, build trust and confidence with each participant, ensure each participant has a positive experience throughout the health assessment process and ensure respect and acknowledgement of each participant. Feedback from the pilot study and from clinical experience informed the researcher that this type of approach would allow the participants the opportunity to fully engage in the process. Therefore from the time contact is initiated until the participant completes their assessments the assessor will endeavour to ensure a positive experience through the principles of the emancipatory philosophy.

- Social engagement
- Reciprocity
- Gain
- Empowerment

The way the room is set up, the way the person is greeted, the way the assessments are administered, the clear plain information and explanations given, being conscious of one's own body language at every juncture of the appointment respects the participant and ensures choice of action. In that way the assessment process is a more active shared engagement with the participants than the participant being a passive recipient. In summary, making reasonable adjustment to the process through the key fundamentals of emancipatory philosophy to ensure each participant can be the best they can be on the day.

2.3 Operationalising reasonable adjustment

Introduction

Specific conditions and adjustments are required to ensure a successful and enjoyable experience for all participants. These adjustments to the objective health measures are viewed as necessary and beneficial to all stakeholders. As well as the development of the accessible explanatory material the following points portray the adjustments the assessor instigated from her own experience and following consultation with independent advocates and advocacy groups, experts in the field of ID and the Scientific Advisory Board to the IDS-TILDA study.

1. What the assessments should be called to reduce potential for negative connotations:
   To avoid the possibility of the participants perceiving themselves as being tested and to reduce the potential for participants not giving themselves the opportunity to attend the health assessments due to previous poor experiences with health tests or health examinations, the assessor changed the name of the objective assessments to Health Fair©.

2. How the person to be included in the study was contacted:
   The first point of contact was through poster advertisement. The assessor provided each of the main study field researchers with posters advertising the Health Fair© and advising participants that they would be contacted shortly. For efficiency the assessor contacted the service provider link person to assist with the initial notification of the participants. The assessor then agreed a timeframe and venue for the assessments to take place for each of the services.

3. Where the assessments would take place:
   The health assessor identified the mammoth task of providing appointments for participants at a time and venue suitable to their individual desires similar to the main face-to-face interview. This would be logistically time consuming, very costly and difficulties regards transporting and setting up the equipment in houses had been identified through the pilot study by the assessor, who was a lone researcher. However the assessor wished to ensure the experience was individual. Working through the service provider's link person the appointments were arranged within a specific timeframe and forwarded to the link person who liaised with the participants and adjusted the appointments accordingly to suit. Once the appointments (date and time) for that service were agreed the link person returned the list to the assessor who completed individual appointment cards and created information packs for each of the participants. The appointments and information were posted to the individual participants a minimum of one week prior to the appointment. The appointment card had the assessor's photo and contact details. This allowed the participant time to contact should they or their support person have any queries and to know who was coming to do the Health Fair© with them.

4. The process of meeting and creating a welcoming, relaxing and respectful atmosphere:

   On the day of the health assessments the assessor ensured sufficient time was allotted to both the setting up and checking of the equipment and for each of the appointments. Previous timing during the pilot study had indicated that in total 30minutes were sufficient for completion. The health assessor allowed 5 – 10 minutes contingency for people who maybe slower or require more time to understand the process. The assessor met each participant with a welcoming greeting at the door of the Health Fair© room, invited them to come in and used a number of different ice breakers to develop rapport and relaxation. All equipment and documentation was pre-prepared so no distraction could ensue, the participant received full attention. If a support person was accompanied the participant they were equally shown respect and greeted, however it was made clear that all conversation would be directed to the participant no matter what level of communication ability the participant held. However, the support and direction of the support person was welcomed and accepted.

5. The method of gaining consent:

   Gaining consent from some people with intellectual disability can prove difficult. Prior knowledge through discussion with the link person was helpful in establishing the level of understanding of the participant. The support person assisted the assessor understand those whose diction was unfamiliar to the assessor. At all times the assessor observed body language and followed protocol as with point 3.1 above. The assessor always sat on a small stool and could therefore ensure she was always in the line of sight of the participant.

6. The behaviour of the researcher to reduce overpowering or standing over the person, give clear instruction and demonstration, undertake assessments only when the person was happy to do so:

   Utilising the small stool enabled the reduction of the assessor being overpowering/standing over the person. The accessible material enabled clear instruction and understanding. The introduction of alternative methods of communication (Lámh, hand over hand, picture representations) and demonstrations promoted good understanding of the assessments. No assessment was undertaken

unless the participant was completely happy to do so. The support person assisted with informing the assessor on the various indicators participants with no verbal language possessed and their meaning, for example particular sounds or vocalisations when they were happy with something.

7. The way the measures were administered:

   No measure was undertaken without the participant being happy to do so, at all times the assessor allowed the participant to take the lead. Some of the measures proved difficult for some participants. Specific techniques were developed to overcome difficulties identified. If these techniques were not suitable alternate measures, where available, were offered. The following table presents the difficulties identified and the methods employed to overcome these difficulties. Each of these methods has been built into the Health Fair© protocol.

Reasonable adjustments employed to difficulties identified	
Difficulties identified	Adjustments method employed
Mounting the standiometer – the participant became anxious, approached the standiometer face on, grabbed at the pole of the standiometer increasing the potential of falling or breaking the equipment.	• Position participant beside standiometer with their back to the wall • Assessor stands directly in front of participant • Show the participant the feet prints on the standiometer by pointing to them • Tell the participant they are going to do exactly what the assessor is going to do • The assessor stretches out their hands and instructs participant to hold both hands with their hands (this prevents the participant grabbing the pole and also provides stability for the participant) • The assessor continuously encourages the participant that they are doing well, to allay fear and anxiety • The assessor instructs the participant to take a step sideways onto the feet prints on the standiometer, the assessor steps sides simultaneously • The participant is now ready to have the measurement complete • Follow instructions as before regards breathing technique and sliding indicator

Stepping onto weighing scale – anxiety and nervousness demonstrated when attempting to step onto scales perhaps due to the possibility of missing scales due to vision impairment	• Instruct the participant that you are going to weight them now • Prepare for weighting – no heavy clothes, no shoes • Position scales with sufficient space that the participant can stand comfortably in front of the scales with the assessor directly opposite • Stand participant beside scales and have them hold the assessors hands • Get them to take a deep relaxing breath in and out • Instruct the participant to take a step up and forward onto the scales using the assessor for balance whilst they are mounting the scales • Where floor covering was dark similar to the scales top the assessor placed a square of brightly coloured linoleum beneath the scales.
Applying isopropanol solution – the solution never reaches room temperature and therefore feels very cold to a warm foot. The sudden shock from the cold is uncomfortable for the participant and can cause the participant to withdraw their foot from the assessment	• Ensure the QUS is ready to receive the participants foot i.e. all necessary information has been keyed in and the forward button has been set to the required point • Ensure the participant is ready shoe and sock off and foot resting on side of the QUS • The assessor sprays the required amount of isopropanol into the palm of their hand • Letting the participant know the assessor gently takes the participants foot to place it into the machine and align on the foot plate correctly • Simultaneously rub the hand containing the solution in a circular motion around the back of the foot spreading the solution on the total heel • Depress the button to commence the measurement immediately all the time letting the participant know what is happening and providing them with encouragement
Distress when automated BP monitor became tight – although one of the most familiar assessments the automated BP monitor inflates to nearly 200mmHg which causes discomfort and sometimes	• Employ distraction technique to reduce distress • Inform the participant that the machine does get tight around their arm • Tell them that you will let them know

distress for participants	when it is going to get tight • When the numbers begin to rise tell the participant to watch the numbers going up • Once it reaches 100mmHg tell the participant that it's going to get tight now and they should take a deep breath • Get the participant to breath in deeply with the assessor demonstrating at the same time • Tell the participant to blow it all out gently demonstrating and encouraging the participant at all times
Poor understanding of the concept of squeeze – The assessor deemed this assessment as one of the most difficult, obtaining grip strength posed a difficulty as the participant did not get any feedback or effect from doing the assessment, the measuring dial is facing away from the participants observation and the squeeze action yields no discernible movement of the handle that is held	• Demonstrate the equipment to the participant pointing out the hands on the clock and how they move • A soft rubber ball is used to demonstrate the squeeze action • Instruct the participant to squeeze the ball • Ensure the handle of the Jamar is correctly positioned and instruct the participant to squeeze the way they squeezed the rubber ball • Use hand over hand to demonstrate the action and feeling of squeeze • Encourage the participant even if the measurement was not a success
Tape on floor viewed as obstacle – some participants stopped short of the tape, walked around it or attempted to step or jump over it	• Demonstrate the TUG assessment • Point out the tape clearly noting that it is not an obstacle • Feel the tape with your foot or hand demonstrating to the participant that it is not raised from the ground • Allow the participant to feel the tape or touch it with their foot or hand

8. Developing a shared understanding of the importance of the activity:

It was imperative that the participants were aware of their contribution to both the study and to the future of service development for people with ID. This was included in the introduction to the Health Fair© and explanation of the study. Further, following completion of their Health Fair© each participant received an individually created certificate of achievement no matter how many of the assessments they had completed.

9 Adjustments of the research data collection methods:

Initially position for all the assessments were built into the computer assisted programme for interviewing. This method builds the dataset as the results are entered. However the assessor found that by using the laptop for data entry the flow of the process was broken; the participant at times became distracted focusing on the computer and the assessor was breaking their attention from the participant to the computer. The assessor decided to develop a recording sheet in lieu of the CAPI system which could be completed discretely throughout the assessment process and although this added data entry to the whole process the assessor felt that her attention was not distracted and neither were the participants.

3. Introducing the Health Assessment on the Day

The assessor will have introduced the health assessment by phone and in writing before the day of the assessment. However best explanations are in person the day of the visit. On the day of arrival to the Health Fair© site, introduce yourself to the gatekeeper and if opportunity arises prior to assessments meet participants. Each participant will have received an individual appointment card with their appointment time, venue and picture of the assessor. The picture enables the participant to be familiar with the assessor before they arrives onsite. Ensure you have sufficient time to set up, calibrate and quality check each piece of equipment. Ensure all necessary documentation is ready and in order of appointments.

Requirements

- Participant PIN
- Participant age
- Assessment recording sheet
- Easy read information booklet
- Easy read consent
- Easy read individual assessment explanatory showcards
- Certificate of achievement

At the beginning of the physical assessment before any measurements are taken you will need to explain what you hope to do during the visit and reassure nervous participants that every stage is optional.

3.1 Consent

OBTAIN WRITTEN CONSENT BEFORE COMMENCING ANY ASSESSMENTS

OBTAIN VERBAL CONSENT (AFFIRMATION) TO CONDUCT ASSESSMENT BEFORE ATTEMPTING EACH ASSESSMENT

Consent to participant will be obtained from all participants once full explanation has been given, the participant understands the process and all questions are answered. For those who do not have verbal language ensure support person accompanies them and is present to support you with communicating, ascertain method of communication and observe body language and facial expression closely throughout the Health Fair©. Explain the consent process in plain, clear language – the following is a sample of how you can phrase seeking consent.

> "I would like to start the measures now. Can I have your permission to do the measures? (Pause for affirmation, then reaffirm) You do not have to do them if you do not want to, is this OK? (Pause)Will you sign the consent form? By signing this you are allowing me do the measures with you. We can stop at any time, is that OK? (Pause) We will do one at a time. You can let me know how you feel about them".

It is very important that no measurements are attempted until the consent has been obtained. Utilising the easy read consent form confirm with the participant that they understand what is going to happen, why the measures are being taken and that it is completely voluntary to participate. Obtain written consent either signature or the participants mark. The mark ought to be witnessed by the support person and a note made regards the fact the participant gave agreement with a mark. Where the participant is unable to sign or make a mark their verbal assent is acceptable and ought to be also witnessed by the support person.

3.2 Feedback

Participants will be given a copy of the following results on the day of their home assessment

- Height
- Weight
- Blood Pressure
- Heel Ultrasound

Participants must agree and/or specify if they want a copy of their results to be given to their carers or key workers.

3.3 Address Health Abnormalities

It is important to remember that IDS-TILDA is primarily a research study and we are not offering a health check. However some of the measures are of known clinical importance and serious abnormalities cannot be overlooked. If a cause for medical concern is identified during the health assessments it is important to follow procedures as per field researcher's protocol in order to deal with this effectively

If the participant's blood pressure is above 140/90 mmHg, link with the participant's carer/key worker as they may have a known blood pressure abnormality and be on medication for hypertension. If not they should be advised to see their own doctor as soon as possible to discuss. If it is substantially above that (>200 mmHg systolic or > 110mmHg diastolic) or if there is any other cause for concern, notify their carer/keyworker and the advice should be to visit their doctor without delay (either their GP or the nearest accident and emergency department). It is important that the respondent understands that their GP is not involved in the IDS-TILDA study. Therefore any visit(s) to their GP are the participant's responsibility and will be in line with the usual arrangements they and their GP have.

3.4 Addressing Participant Distress

If the participant has let you know that they want to stop the physical examination due to distress, or you feel that the physical examination should be stopped let the person talk, and be willing to listen to whatever they have to say. Be empathic. Ask them what they think they might do to feel better. Do they have someone they could talk to for example their keyworker or carer?

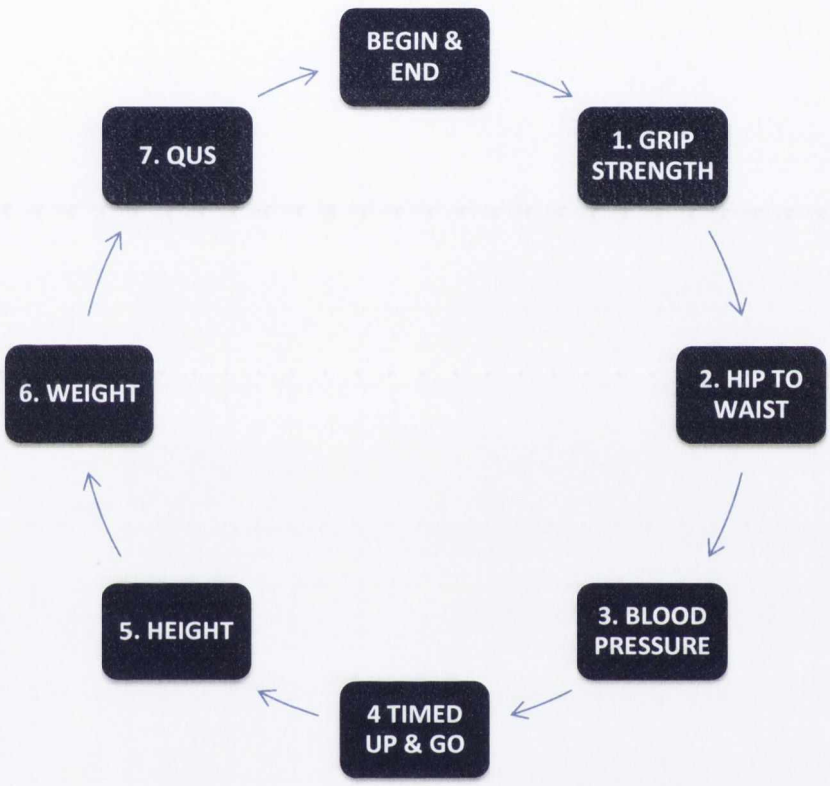
If alone, do they have /know their keyworkers/carers number they/you could call? Who would be a source of strength and reassurance to them? Alternatively, seek assistance from their key worker or carer.

- Try to allay their fears and anxieties about the assessments.
- Resume the physical examination or offer an option of rescheduling, **if appropriate**.

- Depending on the reason for distress, you may deem it more appropriate **NOT** to resume the assessment at another time.

4. The Health Fair© Assessments

Following pilot testing the following order for each health assessment has been deemed the least burdensome



Each measurement is purposely positioned to reduce the stress and therefore impact on the assessment reading, also to minimise inconvenience and burden for the participant.

NOTE: Blue text bold and in inverted commas indicates sections that should be read to the participant

4.1 Grip Strength

Introduction

Hand-grip strength affects every day function and declines with age. It is an indicator of frailty in older persons and lower grip strength is associated with higher morbidity and mortality.

Equipment

- Rubber ball
- Grip strength is measured with a BASELINE HYDRAULIC HAND DYNAMOMETER which consists of a gripping handle with a strain-gauge and an analogue reading scale (Figure 1)

Figure 1:
Max indicator



DUE TO DIFFICULTY ASCERTAINING DOMINANCE MEASURES WILL BE TAKEN ON BOTH HANDS IF POSSIBLE. RECORD ANY ABNORMALITIES OBSERVED

Exclusion criteria

Those with swelling or inflammation in their hand/wrist, severe pain or recent injury to their hand/wrist, and those with surgery to their hand/wrist in the last 6 months should not take the grip strength test. If there is a problem with one of the participant's hands, just take measurements on the other hand. Record all abnormalities observed.

Demonstrate

Firstly demonstrate the motion of squeezing using the rubber ball. Then demonstrate the grip-strength test for the participant. It is very important that you demonstrate the measurement correctly. Experience has shown that participants follow more closely what the interviewer does rather than what s/he says. If the participant indicates that s/he does not understand how to handle the dynamometer, demonstrate it again rather than relying on repeating verbal instructions.

Accuracy

The accuracy of the tests depends on the effort exerted by the participant and the conscientiousness of the interviewer. Consequently it is crucial that the examination protocol be observed painstakingly. Insufficient effort on the part of the participant will cause the test results to be inadequate for analysis.

Procedure: PLEASE REFER TO ACCESSIBLE INFORMATION LAMINATED CARD TO ASSIST WITH EXPLANATION.

1. Explain the procedure using the accessible material.
2. Show participant dynamometer
3. Show the participant the clock feature and how the hands move when squeezing the machine.
4. Ascertain dominant hand (confirm if possible previous observation from signing consent)- ask the participant which is their dominant hand, if they have difficulty with this concept ask which is their strongest hand. If participant is non-verbal ask them to squeeze your fingers. Record answer on record sheet.
5. The participant should sit in an upright chair.
6. Large rings may need to be removed.
7. Explain and demonstrate the motion of squeezing using the rubber ball stating "**I am going to get you to squeeze the machine like this" (squeeze ball as demonstration).**
8. Tell the participant they will move the hands of the clock by squeezing
9. Demonstrate the test procedure with the dynamometer.
10. **VERY IMPORTANT:** Set handle to comfortable grip for the participant. To do this loosen handle with black rubber adjuster attached to the lower section of the

dynamometer and move the handle to an outer ridge for somebody with a large hand. Move the handle to an inner ridge for someone with a smaller hand. If it is in the right position the metal bar (the grip) should rest on the middle piece of the four fingers.

11. Make sure the participant keeps their upper arm tight against their trunk.
12. The forearm should be at a right angle to the upper arm. If the participant is finding the dynamometer too heavy to hold it in this position then the assessor can use their hand to rest the dynamometer on.
13. The dial should face outward (away from the participant).
14. Allow the participant to handle the dynamometer and become comfortable with the correct positioning.
15. Once you are ready to begin, re-set the max indicator (the red arrow) to zero by turning the white knob anticlockwise.
16. Ask the participant to squeeze the handle with maximum force. **"I'd like you to squeeze as hard as you can for a few seconds like you did with the rubber ball".**
17. Before each measurement, make sure that the max indicator (i.e. red arrow) is set to zero.
18. **Record the value to the nearest whole number (odd or even) in Kg's** (e.g. 22kg or 23kg). To get an accurate reading and avoid the introduction of parallax error (A parallax error is the perceived shift in an object's position as it is viewed from different angles). A guide for doing this is to look directly at the scale holding the dynamometer at a 90 degree angle directly out from your body and positioning the white button at the centre of the dial in line with your nose. Note the scale increases in increments of 2 kg's (i.e. there are only 4 graduations between 20 and 30 kg – these represent 22, 24, 26 and 28 kgs). If the dial falls between 2 graduations, record this number (i.e. if the dial falls between 22 and 24, record the grip strength as 23kg and not as 22 or 24kg).
19. Record two values with each hand alternating between hands, starting with the non-dominant hand (four values all together).
20. Do not include measurements carried out incorrectly.

4.2. Blood Pressure

Introduction

The blood pressure is measured in terms of millimetres of mercury (mmHg). For this test the blood pressure will be measured using an automated sphygmomanometer (Figure 2).



Figure 2 Omron Blood Pressure Monitor

A person's blood pressure should be below 120 over 80 (120/80). This is the ideal blood pressure for people wishing to have good health and at this level there is a much lower risk of heart disease or stroke.

To take the participants blood pressure reading, ensure they are relaxed and comfortably seated, with their arm well supported. Measuring your blood pressure is best done after you rest for at least 5 minutes it will therefore not be the first assessment following commencement of the assessments. Sufficient time is necessary to allow the participant to become comfortable with the process, you the assessor and to assume a relaxed state.

Do not take the participants blood pressure if they have consumed caffeine or used a tobacco product in the past 30 minutes, or have recently exercised.

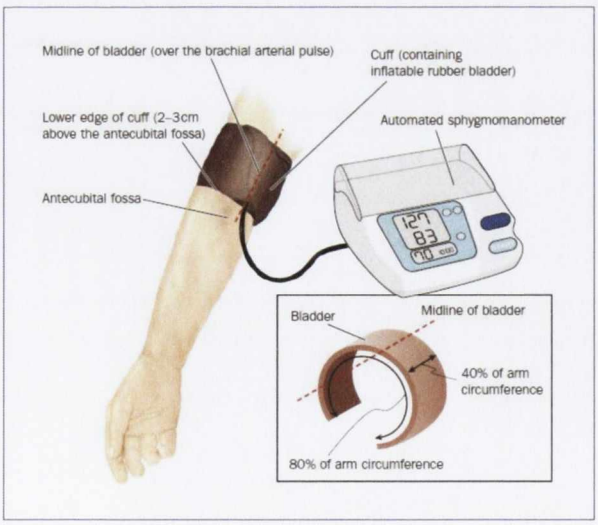


Figure 3: positioning the blood pressure cuff.

Equipment

- Sphygmomanometer
- Chair

Procedure: PLEASE REFER TO ACCESSIBLE INFORMATION LAMINATED CARD TO ASSIST WITH EXPLANATION.

1. Inform the participant you are going to take their blood pressure, **"I am now going to take your blood pressure"**
2. Explain the procedure to the participant using the accessible material.
3. Explain that there will be 4 measurements in total, 2 sitting down and 2 standing up.
4. You will tell them when you are going to do the standing blood pressure.
5. You will count down like this 5, 4, 3, 2, 1. When the participant hears 1 they will stand up with their arms by their side.
6. Between each blood pressure there will be a short rest of one minute.
7. To begin the assessment make sure the participant is sitting relaxed with their upper arm at heart level, their back supported, their legs uncrossed and feet on the floor.
8. Ask the participant to expose their upper arm, ensure their sleeve is comfortably rolled up and there are no tight clothing on the arm. "**I would like to roll up your sleeve and place the blood pressure cuff on your upper arm" (if rolling up the garment cause bulkiness do not do this once there are no restrictions a light garment can be left in place)**
9. Turn on the machine and position it as above on a level surface.
10. Wrap the cuff snugly around the upper arm.
11. Inform the participant they will feel tightness around their arm. Employ distraction and relaxation technique to allay anxiety or discomfort. **"When I start the blood pressure machine the cuff will get tight but that will only be for a few seconds. Watch the numbers and when they get big (or let them know) we will take a deep breath in and then breath out gently"**
12. Begin the measurement
13. Take the reading when the automated sphygmomanometer indicates the each assessment is complete.
14. Record the each reading on the record sheet.
15. Watch the timer for the third reading when it comes to 8 tell the participant that you are about to do the countdown for them to stand when they hear 1. **"I am about to start counting please stand up when you hear 1"**
16. At 5 begin the countdown **'5, 4, 3, 2, 1 Please stand up (repeat following one minute rest).**

17. Take the readings and record on the record sheet

For those who are unable to stand or are too unsteady to stand independently record sitting blood pressure only and note in record sheet reason for not taking standing blood pressure

4.3 Timed Up and Go (TUG)

Introduction

The timed "Up & Go" test measures, in seconds, the time taken by an individual to stand up from a standard arm chair (approximate seat height of 46 cm, arm height 65 cm), walk a distance of 3 meters (approximately 10 feet), turn, walk back to the chair, and sit down again.

The participant wears his/her regular footwear. If the participant usually uses an assistive device such as a cane or walker, he/she can use it during the test. **No physical assistance should be given during the test.** For those who are unable to participate in this assessment record rationale on record sheet (e.g. non mobile, wheelchair, injury, un-cooperative)

Equipment

- Arm chair (standard chair 45 cms used throughout Health Fair©)
- Tape measure
- High visible tape
- Stop watch.

Setting Up the test area

- Determine a path free from obstruction
- Place the chair with arms at one end of the path. The chair should be stable and positioned such that it will not move when the participant moves from sitting to standing (i.e. against the wall).
- Using high a measuring tape measure mark off a 3 m (10 ft.) distance from the front leg of the chair, mark the distance with high visible tape. Place a strip of high visibility tape on the floor at the foot of the chair also, this will ensure accuracy of the course should the participant move the chair.
- Record the type of floor covering on the record sheet.

Procedure: PLEASE REFER TO ACCESSIBLE INFORMATION LAMINATED CARD TO ASSIST WITH EXPLANATION.

1. Explain the procedure using the accessible material
2. **"Now I am going to do a walking assessment with you. I will get you to sit in this chair with your back resting against the back of the chair. On the word GO you should stand up, walk to the line on the floor, turn around, walk back to the chair and sit down. Please walk at your regular pace. Is that OK? Do you have any questions?"**
3. **"I will demonstrate this now".**
4. Demonstrate the TUG once. Ensure you have given the full instruction prior to the demonstration. Do not talk during the demonstration.
5. Touch the floor tape to demonstrate to the participant that this is not an obstacle.
6. **"I will now get you to do that. Do you have any questions before we begin?"**
7. Ask the participant to sit in the chair. Ensure that the chair is stable and positioned such that it will not move when the participant stands up.
8. **"I will now get you to do a practice run. When I say GO, you should stand up, walk to the line on the floor, turn around, walk back to the chair and sit down. Please walk at your regular pace. Is that OK? Do you have any questions?"** If they do, repeat the instructions.
9. **"GO"**
10. After the practice run, allow the participant to sit down again in the chair. If the participant has problems with the test, you should explain the instructions again but do not demonstrate the task or allow them any more practice runs.
11. It is important to ensure that the pace the participant walked at during the practice was indeed their usual pace and not a pace based on your demonstration of the task. Enquire **"was that your usual pace?"** If so, continue to the timed task. If not, it is important to re-iterate that they need to walk at their usual pace for the next part of the test.
12. **"We are now going to repeat the test but this time I am going to time you. You should walk at your usual pace (as before). Are you ready? GO"**
13. Start timing on the word "GO" and stop timing when the participant is seated again correctly in the chair with their back resting on the back of the chair.
14. Ensure to give encouragement to the participant **"Well done, you are doing great"**

15. Observe participant carefully to ascertain if they require instruction throughout the assessment, provide direction if necessary **"Yes, all the way to the line, now turn around, walk all the way back, well done"**
16. Record the result, to 2 decimal places on the record sheet.

The participant wears their regular footwear, may use any gait aid that they normally use during ambulation, but may not be assisted by another person. There is no time limit. They may stop and rest (but not sit down) if they need to.

Normal healthy elderly usually complete the task in ten seconds or less. Very frail or weak elderly with poor mobility may take 2 minutes or more.

4.4 Anthropometric Measurements

Introduction

Anthropometric measurements include height and weight as well as waist and hip ratio. Body Mass index (BMI) is simply a ratio of weight in relation to height.

$$\text{BMI} = \frac{(\text{weight in kilograms})}{\text{height in meters}^2}$$

BMI is not a direct measurement of body fat, and importantly this ratio provides no information about distribution of body fat. Conversely the waist-to-hip ratio is a measure of distribution of body fat both subcutaneous and intra-abdominal. Studies suggest that this ratio is a predictor of health risk similar to that identified by BMI. For those who cannot stand independently or for some other reason height, weight or height and weight are unable to be taken please use alternate measures in accordance with the MUST tool. For height – Ulna length and for BMI (weight) MUAC will be utilised.

4.4.1 Height

Introduction

Height will be measured to the nearest millimetre using a free-standing stadiometer or height measuring device (figure 4). **Assemble the stadiometer against a wall ensuring adequate space, no obstruction or risk to participant (pre-assembled on arrival to**



Figure 4 Secca Standiometer

Health Fair© site). The height will be used in the calculation of the participants Body Mass Index (BMI).

> **TIP:** For ease of assembly have each piece of the standiometer pole marked in order of connection.

Equipment

- Portable Stadiometer

Procedure: PLEASE REFER TO ACCESSIBLE INFORMATION LAMINATED CARD TO ASSIST WITH EXPLANATION.

1. Using accessible material explain the procedure to the participant.
2. Inform the participant. **'I am now going to measure your height'.**
3. Ask them to remove their shoes and stand at the stadiometer. **'Please remove your shoes and stand beside the height measuring device with your back to the wall'.**
4. Ask the participant to hold your hands and take a step sideways onto the feet on the standiometer, you can demonstrate as they are doing it too. **"Please hold my hands for steadiness and take a step onto the feet on the measuring device"**
5. Ask the participant to look straight ahead, stand still, take a deep breath, let it out. **"Please stand as still as you can, look straight ahead, take a deep breath and let it out gently".**
6. Bring the slider down until it is resting on the participants head.
7. Take a reading from the stadiometer and record in centimetres on the record sheet
8. Assist participant to the weighing scales.

4.4.2 Weight

Introduction

Weight will be measured to calculate the participants BMI. Weight is measured to the nearest 100 g using Ultra slim digital Secca scales. Turn on the scales. Request the participant to remove their shoes before the measurement is undertaken (if not already removed) and any

extra or heavy clothing. Request the participant to stand on the scales. Assist them to step on if necessary. Take reading and record in kgs on record sheet.

Equipment

Ultra slim digital Secca weighing scales

Procedure: PLEASE REFER TO ACCESSIBLE INFORMATION LAMINATED CARD TO ASSIST WITH EXPLANATION.

1. Inform the participant. '**I am now going to measure your weight'**
2. Turn on the scales.
3. Ask them to stand on the scales. **'Please remove your shoes and stand on the weighing scales'.**
4. Support the participant as they step onto the scale by standing directly in front of them and with their hands resting on your hands
5. Tell participant you are going to let go your hands
6. Ask the participant to stand as still as possible. '**Please stand as still as possible'**
7. Take a reading from the scales and record in kilograms on the record sheet.

4.4.3 Waist Circumference

Introduction

Regardless of height and build, a relative excess of fat around the waist appears to increase the risk of developing one or more chronic diseases. One way doctors recommend assessing your weight-related risk is by measuring your waist circumference in relation to your hip circumference. The measurement of waist circumference provides information regarding fat topography - where body fat is stored. This is important because people who carry their weight centrally are particularly at risk of developing heart disease and type 2 diabetes. When measuring waist circumference it is important not to be tempted to measure around the narrower part of the abdomen situated below the umbilicus. Using the anatomical landmark of the iliac crest and ensuring that measurement is taken on a horizontal plane just above this level provides the most accurate, reliable and reproducible technique for waist circumference measurement (see Figure 5)

Equipment

SECA measuring tape

Exclusion

- Those who cannot stand or are unable to lie flat e.g. wheelchair bound
- Those who have a physical compromise such as colostomy or ileostomy
- If there are any other reasons why the measurement was not taken record.

Preparing the Participant

If the participant is wearing outer layers of clothing without embarrassing them request the removal of these outer layers. Explain that you will be touching their hip to assist with locating the correct position for the tape. Explain to the participant the importance of the measurement and that the layers of clothing can substantially affect the reading.
Where possible

- Remove heavy layers of clothing
- Remove shoes with heels
- Tight garments that would alter the shape of the body.

Ensure the participant is standing erect in a relaxed manner and breathing normally. Weight should be evenly balanced on both feet and the feet should be about one foot (30cms) apart. The arms should be hanging loosely at their sides. Sit or stand at to one side of the participant.

Using the measuring tape

Ask the participant to hold the end of the tape measure. Pass the tape around the body of the participant. Hook the tape end with lock into the main body of the tape. Press button for ease of adjustment of tape measure. To check the tape is horizontal you have to position the tape on the right flank and peer round the participants back from his or her left flank to check that it is level. This will be easier if you are kneeling or sitting on a chair to the side of the participant. Hold the tape flat against the body and flatten the end of the tape to read the measurement from the outer edge of the buckle. Do not pull the tape towards you as this will lift away from the participant's body, affecting the measurement.

Please note that you must enter the measurement to one decimal place – do not round it to the nearest centimetre. All measurements should be taken to the nearest millimetre – for example 78.4 cms NOT 78 cms.

Locating the waist

The waist is defined as the point midway between the iliac crest and the costal margin (lower rib). To locate the levels of the costal margin and the iliac crest use the fingers of the right hand held straight and pointing in front of the respondent to slide upward over the iliac crest. Men's waists tend to be above the top of their trousers whereas women's waists are often under the waistband of their trousers or skirts.

Accurate measurement of waist and hip circumference is achieved using the following technique:

Procedure: PLEASE REFER TO ACCESSIBLE INFORMATION LAMINATED CARD TO ASSIST WITH EXPLANATION.

1. Using the accessible material explain the procedure to the participant.
2. Inform the participant you are going to measure their waist. "**Now I am going to measure your waist and hips. I have to place the tape in the correct position".**
3. Demonstrate where you are going to place the tape. "**I am going to place my hand on your hip like this**".
4. Ensure the participant agrees and is comfortable with having their measurement taken.
5. Locate the top of the hip bone (iliac crest) and take the measurement just above this bony landmark, just where two finger can fit between the iliac crest and the lowest rib (Figure 6).

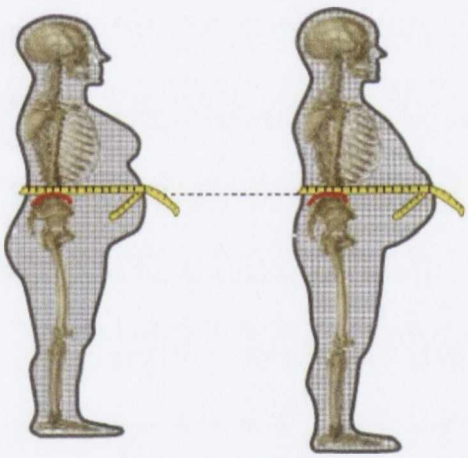


Figure 6 Identifying waist positioning

6. Ensure that the tape measure is positioned horizontally, parallel to the floor.
7. Measuring at a level just above the iliac crest, and positioning the tape horizontally, irrespective of whether the umbilicus is above or below the tape, provides the correct waist circumference measurement and should correspond to the maximal abdominal diameter.
8. Ensure that the participant is standing erect and has relaxed the abdominal muscles. Measurement is taken at the end of normal expiration.

9. If participant cannot stand request they lie down flat and undertake the measurement.
10. Aim to have a snug but not too tight a fit of the tape measure around the waist; do not make compressions in the skin with the tape measure.
11. Record the waist measurement in centimetres on the records sheet.

4.4.4 Hip Circumference

The hip circumference is defined as being the widest circumference over the buttocks and below the iliac crest. To obtain an accurate measurement you should measure the circumference at several positions and record the widest circumference. Four measures are recommended if possible.

1. Check the tape is horizontal and the respondent is not contracting the gluteal muscles. Pull the tape, allowing it to maintain its position but not to cause indentation. Take 2 quick measurements of the hips at the widest point and record the largest value on the recording sheet
2. Take a further waist measurement and record on record sheet.
3. Following the second waist measurement take a further 2 quick measurements of the hips at the widest point and record the largest value on the record sheet.

Order of measurements

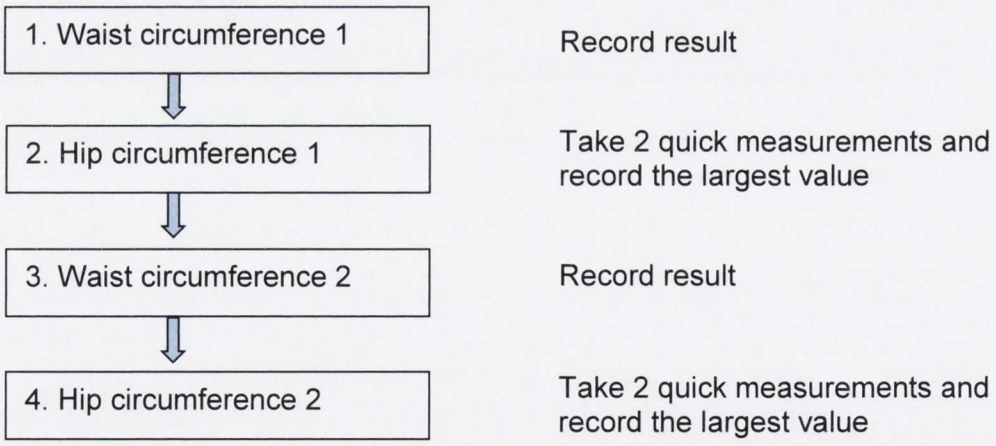


Figure 7 Waist to hip order of measurement

Important to Remember

- The tape should be tight enough so that it does not slip but not tight enough to indent clothing. If clothing is baggy, it should be folded before the measure is taken.
- If the respondent is large, ask him/her to pass the tape around rather than having to "hug" them. Remember though to check that the tape is correctly placed for the measurement being taken and that the tape is horizontal all the way around.
- If your second waist or hip measurement differs by 3cms or more from the first, you should take a third measurement.

4.5 Quantitative Heel Ultrasound (QUS)

Plug in the machine and switch on - the machine needs to be plugged in about half an hour before you want to use it, to give it time to warm up it is therefore efficient if you have done this at the beginning of the health assessment.

Introduction

Osteoporosis is a disease of bone leading to an increased risk of fracture. Osteoporosis is most common in women after the menopause, but can also develop in men. It can be prevented with lifestyle advice and medication, and preventing falls in people with known or suspected osteoporosis is known to prevent fractures.

Quantitative assessment of osteoporosis and estimation of fracture risk relies mainly on bone mineral density (BMD) measurements using radiologic methods such as dual-energy X-ray absorptiometry (DXA). The gold standard for measuring bone mineral density is a DXA machine. By the WHO criteria, a T score of -2.5 or higher on a DXA indicates osteoporosis. However DXA machines are expensive, large and involve ionising radiation.



Figure 8 QUS foot positioning between inflating balloons

For the purposes of this study, quantitative ultrasound (QUS) will be used for measuring bone density. QUS is a technology for measuring properties of bone at peripheral skeletal sites, is more portable and less expensive than DXA, without the use of ionizing radiation. The site for the measurement will be the participant's heel. The device measures broadband ultrasound attenuation (BUA; expressed in dB/MHz) and speed of sound (SOS; expressed in m/s). BUA is the rate of attenuation in the acoustic energy across a broad range of frequency and this attenuation is due to absorption and scattering of ultrasound in the bone and soft tissue. BUA is influenced by both density and structural parameters. SOS is the ratio of propagation distance to the pulse transit. The foot will be placed in the device and positioned between the two temperature controlled balloons that will inflate (See figure 8). For assessment the regular method is to use the non-dominant foot unless contraindicated by pathology such as ankle oedema, trauma or fracture, reflex sympathic dystrophy, in such cases use the dominant foot. However due to the difficulty is ascertaining dominance with people with intellectual disability both feet will be measured and the lower of each of the measures will be obtained as the measurement of choice. All measurements shall be recorded at the time of measurement.

It is important to realise that the t-score generated from a heel ultrasound is not the same as that generated by a DXA machine and cannot be used to diagnose osteoporosis. However,

having a low score on a heel ultrasound is an independent risk factor for hip fracture (the EPIDOS study, a large prospective study with >5000 women).

Equipment

- GE Lunar Achilles EXPII QUS
- 70% Alcohol spray
- Sterilizer wipes

Quality Assessment

At the start of each day, carry out a quality assessment of the machine and record the result in the equipment book

- Press Quality Assurance.
- Remove the cylinder from the body of the machine and take it out of the plastic bag.
- Spray both sides of the cylinder with the alcohol spray and both membranes in the machine.
- Insert the cylinder in the machine, simulating an ankle/heel and press ok.
- The membranes will inflate as the machine goes through a series of quality checks.
- Once you have passed the QA, remove the cylinder and replace in the machine.
- Press BACK UP on the screen and record the QA results in record manual.

TIP: The solution utilised on the heel is isopropanol which is 70% alcohol and will never get to room temperature. In the pilot study participants found the spray and the solution too cold and a shock to the warm foot. This caused sudden reaction, either to pull away or complain loudly and subsequently refused to continue. Although you pre-warned people the other problem to arise was that this delay caused the solution to dry quickly on the skin requiring further application, compounding the problem. To overcome this problem the assessor developed a technique of applying the alcohol gel with her hand just as the foot was being positioned. Spray the appropriate amount into the palm of the hand as the participant is positioning the foot on the side of the QUS; apply in a circular motion as you are repositioning the foot onto the foot plate.

Procedure: PLEASE REFER TO ACCESSIBLE INFORMATION LAMINATED CARD TO ASSIST WITH EXPLANATION.

1. Seat the person in a stable chair without wheels directly in from of the QUS device, in a comfortable upright position.

2. Ensure the calf is resting in alignment so that the middle of the calf rests gently on the calf support.
3. NB resting the calf too heavily on the calf support may cause the heel to move too far forward causing erroneous readings, correct positioning is important for valid measurement results.
4. The calf rest may be removed in certain circumstance where the participant finds it too difficult to position their foot in the foot rest due to body shape or positioning.
5. To avoid erroneous readings due to poor positioning ensure the hip, knee and foot are in alignment, this is achieved by gently tipping the knee inward until alignment is achieved, unless otherwise indicated e.g. participant contractures or restricted or painful movement of either knee or hip.
6. Inform the participant that you are going to do the measurement. Refer to the accessible material as you explain the procedure to the participant. **"I would like to do a simple assessment to see how strong your bones are".**
7. Ensure the participant agrees and is comfortable with having the assessment performed, demonstrate if necessary.
8. Usually the non-dominant foot is the one measured however foot dominance is more difficult to determine in people with intellectual disabilities therefore both feet will be measured and calculate the value of absolute difference.
9. Ask the participant to take their shoe and sock off the foot to be measured first and to rest their foot on the edge of the machine **"Please remove your shoe and sock and rest it on the side of the machine" (repeat for other foot measurement)**
10. Assess the heel for any abrasions, cuts or open wounds – cease measurement if skin is not in a condition to proceed i.e. compromised
11. <u>While the participant is preparing</u>, enter the participant's age, gender and which foot you are measuring. This will expedite the process and avoid delay, especially if the participant is anxious or nervous about the assessment.
12. <u>It is important to have the machine ready to go (i.e. click the screen on to the point for inflating the balloons) prior to placing the foot on the footplate.</u>
13. Click forward: A diagram now appears on screen instructing you where to spray the alcohol gel.
14. Inform the participant you are going to spray their heel with an alcohol spray to help with the procedure and it may be helpful to tell them that it is a bit smelly and cold **OR** use alternate method for application.

15. Spray alcohol gel on both sides of the participant's heel as well as both membranes.
16. Ask the participant to insert their foot into the machine and rest their leg against the leg rest. **'Please place your foot into the measuring device and keep it as still as possible the test takes just 10 seconds.'**
17. Position the foot to ensure the white rib is between the first two toes.
18. Press START.
19. Check and ensure the heel is fully back by running your hand behind the participant's foot.
20. Inform them that they will feel a slight touching sensation on their heel. There is no pressure and it does not hurt or cause discomfort, it is nice and warm. "**You will feel the balloons getting bigger, it is soft and warm, and does not hurt"**
21. Press CONTINUE.
22. The membranes will fill and measurement will start immediately. Two wave forms display on the screen.
23. Make sure the person does not move their foot during the measurement as this can lengthen the measurement time or cause a measurement failure.
24. The membranes deflate after the measurement is completed, remove the foot from the footplate and wipe any remaining alcohol from the foot and membranes.
25. Press print and record the SOS, BUA, stiffness index and t-score on the recording sheets.
26. Repeat for other foot.
27. The assessment is now over. The participant can remove and dry their foot, and put back on their shoes/socks.
28. Complete the participant results card and inform them if they need to show it to their GP/Key worker.

5 Alternate measures

Introduction

The use of the 'Malnutrition Universal Screening Tool' ('MUST') to provide alternate surrogate measures was included for those people who would be unable to engage in assessments such as height and weight measure –mid upper arm circumference (MUAC) for estimation of body mass index (BMI) and ulna length for height. The 'MUST' is reproduced here with the kind permission of BAPEN (British Association for Parenteral and Enteral Nutrition), for further information on 'MUST' see www.bapen.org.uk.

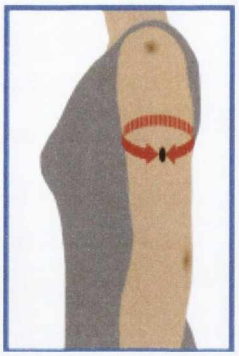


Figure 9 Identifying measuring point

5.1 Mid Upper Arm Circumference

The subject's left arm should be bent at the elbow at a 90 degree angle, with the upper arm held parallel to the side of the body (see figure 9). Measure the distance between the bony protrusion on the shoulder (acromion) and the point of the elbow (olecranon process). Mark the mid-point. Ask the subject to let arm hang loose and measure around the upper arm at the mid-point, making sure that the tape measure is snug but not tight (see figure 10). If MUAC is 32.0 cm, BMI is likely to be >30 kg/m^2. The use of MUAC provides a general indication of BMI and is not designed to generate an actual score for use with 'MUST'. For further information on use of MUAC please refer to The 'MUST' Explanatory Booklet.

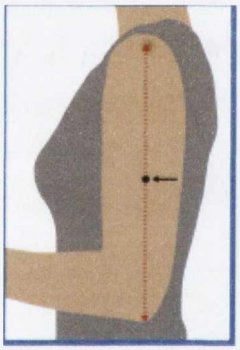


Figure 10 MUAC measuring point

5.2 Ulna Length

Measure between the point of the elbow (olecranon process) and the midpoint of the prominent bone of the wrist (styloid process) (left side if possible). Refer to table for estimation of height (Table 1).

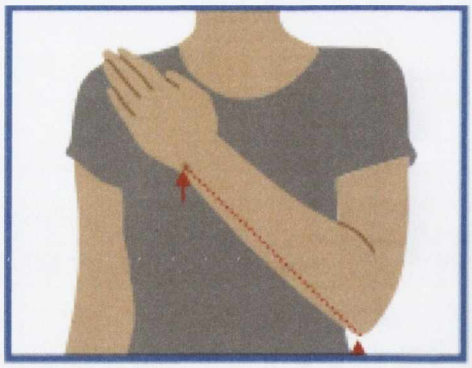


Figure 11 Ulna length measuring point

Table 1: Height estimation calculations from ulna length (cms)

Height (m)	men (<65 years)	1.94	1.93	1.91	1.89	1.87	1.85	1.84	1.82	1.80	1.78	1.76	1.75	1.73	1.71
	men (≥65 years)	1.87	1.86	1.84	1.82	1.81	1.79	1.78	1.76	1.75	1.73	1.71	1.70	1.68	1.67
	Ulna length (cm)	32.0	31.5	31.0	30.5	30.0	29.5	29.0	28.5	28.0	27.5	27.0	26.5	26.0	25.5
Height (m)	Women (<65 years)	1.84	1.83	1.81	1.80	1.79	1.77	1.76	1.75	1.73	1.72	1.70	1.69	1.68	1.66
	Women (≥65 years)	1.84	1.83	1.81	1.79	1.78	1.76	1.75	1.73	1.71	1.70	1.68	1.66	1.65	1.63
Height (m)	men (<65 years)	1.69	1.67	1.66	1.64	1.62	1.60	1.58	1.57	1.55	1.53	1.51	1.49	1.48	1.46
	men (≥65 years)	1.65	1.63	1.62	1.60	1.59	1.57	1.56	1.54	1.52	1.51	1.49	1.48	1.46	1.45
	Ulna length (cm)	25.0	24.5	24.0	23.5	23.0	22.5	22.0	21.5	21.0	20.5	20.0	19.5	19.0	18.5
Height (m)	Women (<65 years)	1.65	1.63	1.62	1.61	1.59	1.58	1.56	1.55	1.54	1.52	1.51	1.50	1.48	1.47
	Women (≥65 years)	1.61	1.60	1.58	1.56	1.55	1.53	1.52	1.50	1.48	1.47	1.45	1.44	1.42	1.40

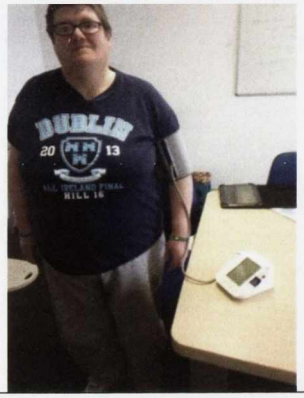
6. Accessible Explanatory Material

The following accessible materials were developed in keeping with the inclusionary ethos of the main IDS-TILDA study. The assessor developed easy-to-read explanatory and supporting material following consultation with an independent advocate, the scientific advisory panel and the PIs of IDS-TILDA. The assessor organised focus group consultations with a number of independent advocacy groups throughout the country. The material was presented to the groups to review and critique. The Health Fair© process and how it would be administered was demonstrated. The groups made a number of suggestions and changes which assisted in improving the overall process. These changes were instigated and reviewed again by the independent advocate.

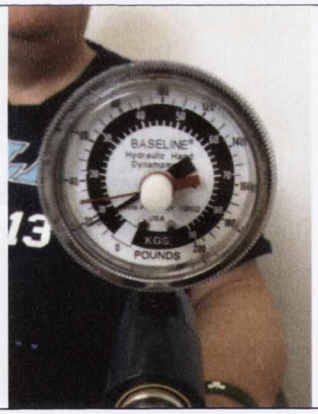
Weight Measurement

	I am going to do your weight measure
	Take off your shoes or boots
	Step on the scales carefully
	Stand still for a few seconds

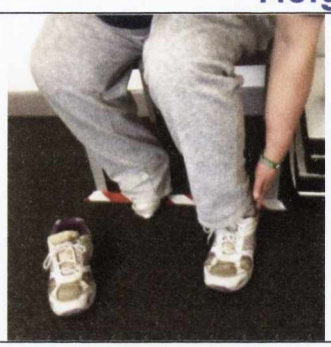
Blood Pressure

	Take off any bulky coat or cardigan
	The cuff will be placed around your arm
	Relax while the machine takes the blood pressure measure First 2 sitting down
40,56,100,134	The numbers will go up and the cuff will get tight only for a few seconds
	For last 2 measures Stand up with your hands by your side when I tell you

GRIP STRENGTH

	Take jewellery or large rings off
 	We will practice the squeeze action like this
	I will show you how the machine works
	Hold the squeeze machine in your hand Squeeze as hard as you can like squeezing the ball

Height Measurement

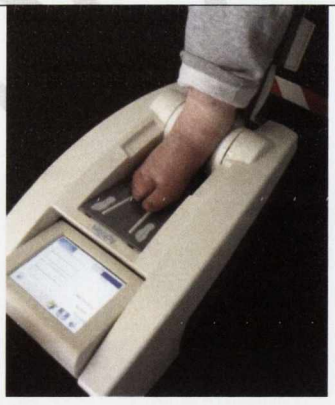
	Take off your shoes or boots
	Stand beside the measuring pole
	Step onto the measuring pole like this
	Look straight ahead. Take a deep breath, breathe out and stand still. I will see how tall you are now

Height Measurement for people who cannot stand

	Place your arm across your chest with your fingers pointing upward
	or Place your arm resting on the table
	Keep still
	You can now relax

PROOF COPY

HEEL ULTRASOUND (QUS)

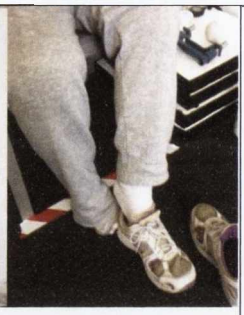
	Remove your shoe and your sock
	I will put your details into the machine Place your foot at the side of the machine
	I will put some spray on the machine
	Sit and relax keeping your foot as still as possible

PHOTOCOPY

TIMED UP AND GO TEST – TUG

	Sit comfortably in your chair
	On the word **GO** stand up 
	Walk **AT YOUR USUAL PACE** to the line on the floor
	Turn around at the line. Walk back to your chair.
	Sit back down

Waist and Hip Measurement

 	Remove your heavy outer clothes and shoes
	Stand up straight Put your arms out like this
	I will put the tape around your waist.
	I will put 2 fingers on your hip I will then measure your waist
	Then your hips

The Intellectual Disability Supplement to TILDA
The University of Dublin, Trinity College,
School of Nursing & Midwifery,
24 D'Olier Street, Dublin 2.

General Inquiries: 01-8963187
Email:eburke1@tcd.ie
Email: idstilda@tcd.ie

www.idstilda.tcd.ie

The Intellectual Disability Supplement to TILDA is funded and supported



Appendix 9

Health Fair Process

FLOW CHART OF CONTACTING, SETTING UP APPOINTMENTS & COMPLETING HEALTH FAIR

IDS-TILDA Main Contact Spreadsheet

Filter by Service Provider

Produce spread sheets per service include Participant name and contact details – maintain within secure folder on R drive in accordance with data protection

Arrange Services with greatest number of participants - in rank order

Develop colour codes to represent contact action

No colour – no action

- Attempted contact nothing confirmed

- Contact confirmed, invited, awaiting confirmation of appointment date and venue. Re-contact within a week if no confirmation affirmed

- Date and venue confirmed move to next stage.

Maintain contact details database Keep notes of contacts on each spreadsheet associated with services

Prepare packs and post appointments

- Create results and recording sheets per participant
- Plan appointment schedule
- Plan route of travel for on the day
- Plot appointments and link person's contacts in diary
- Confirm attendance day before
- Check stocks and equipment before travelling.

Attend appointments and complete Health Fair

- Maintain ethical and data protection principles until raw data is secure in office
- Enter data to secure dataset as soon as is possible and feasible
- On completion of HF move appointment sheet planner to completed folder
- Move spreadsheet planner to end of row

Continue process with service in progress or yet to be contacted

Appendix 10:
Wave 2 Ethics Approval

TRINITY COLLEGE DUBLIN
The University of Dublin
Research Ethics Committee
Faculty of Health Sciences,
Chemistry Building,
Trinity College,
Dublin 2, Ireland.
T:- +353 (0)1 8964255

Professor Mary McCarron
Dean of the Faculty of Health Sciences and Principal Investigator of IDS-TILDA,
Chemistry Building,
Trinity College Dublin,
Dublin 2.

7 February 2013

Study: Intellectual Disability to the Irish Longitudinal Study on Ageing (IDS-TILDA) – Wave 2

Dear Professor McCarron,

Further to the review of the modifications submitted for the IDS-TILDA Study Wave 2, I am pleased to inform you that the continuance of your ethical approval has been re-affirmed by the Faculty of Health Sciences Research Ethics Committee.

Wishing you the best of luck with your study.

Yours sincerely,

pp Cbalfe

Dr. Ruth Pilkington
Chairperson
Faculty Research Ethics Committee

Appendix 11:

Accessible Health Fair Consent Form

Consent Form for Objective Measurements

An Intellectual Disability Supplement
The Irish Longitudinal Study on Ageir

The researcher will explain the measurements to you using the laminated cards.

Please read the information below and sign the consent form if you agree to have the measures taken

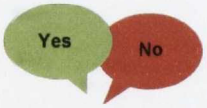
Please tick ☑



I have read the information on the different measures that will be taken ☐



I understand what is involved in taking the measurements ☐



I know that it is my choice to take part ☐

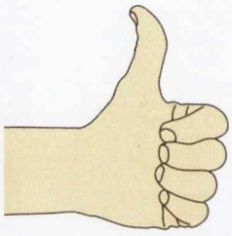


I understand that I can stop whenever I want. I do not have to give a reason ☐

Your consent

Your name:_____

Please tick ☑



I wish to take part ☐

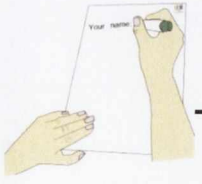
Phone number: _____

Your address:

_____

_____

Please sign or mark your name here:



_____

Date: _____

Carer/Support Person:_____

Appendix 12:
Accessible Health Fair Results Card



An Intellectual Disability Supplement to
The Irish Longitudinal Study on Ageing

THE INTELLECTUAL DISABILITY SUPPLEMENT TO THE IRISH LONGITUDINAL STUDY ON AGEING

HEALTH FAIR RESULTS

My Measurements

Name:

An Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing

	**HEIGHT**	_____ cms
	**WEIGHT**	_____ Kgs
	**BLOOD PRESSURE**	_____ mmHg
	**HEEL ULTRASOUND - QUS**	T Score _____

I should tell my doctor my results

Yes ☐

No ☐

If you have any questions please call



Eilish Burke

IDS-TILDA
Phone: 0872632241 OR 01- 8963187
Email: idstilda@tcd.ie

Intellectual Disability Supplement to TILDA
School of Nursing & Midwifery,
The University of Dublin, Trinity College,
24 D'Olier Street,
Dublin 2,
Ireland.

Email: idstilda@tcd.ie

Website: www.idstilda.tcd.ie













Appendix 13:

Health Fair Invitation Pack

a. Information booklet
b. Accessible cover letter
c. Accessible appointment card



An Intellectual Disability Supplement to
The Irish Longitudinal Study on Ageing

Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing (IDS-TILDA)

HEALTHY HAPPY AGEING

Health Fair Information Booklet

TRINITY COLLEGE DUBLIN
COLÁISTE NA TRÍONÓIDE
THE UNIVERSITY OF DUBLIN



IDS-TILDA

Working to Make Ireland the Best Place to Grow Old

THE HAPPY HEALTHY AGEING HEALTH FAIR

INTRODUCTION



IDS-TILDA is a study about people who are growing older with an intellectual disability in Ireland.



We need you to help us learn more about people's health and how it can change as they grow older.



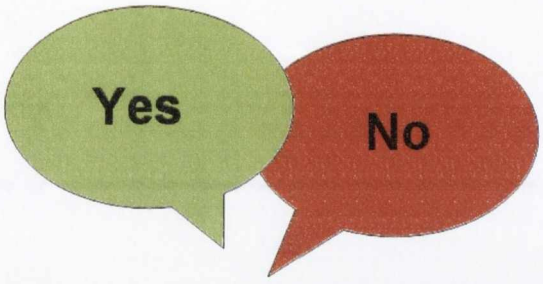
We are going to hold a health fair as part of Wave 2 of the IDS-TILDA study. At the Health Fair we will do some health assessments.



This will help us to learn about people's health as they grow older.



We will first ask for your permission to do the health assessment.



You do not have to take part if you do not want to. It is your choice

GENERAL INFORMATION



None of the assessments will hurt you and you can stop any time you like.

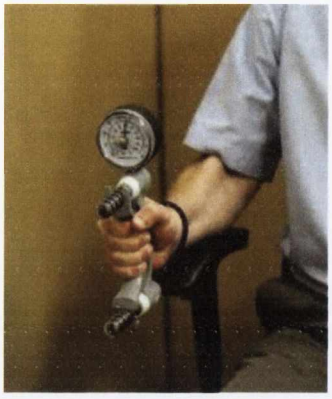


If there are any of the assessments you cannot do that is OK, you can do as many as you are able.



We will arrange the appointment to suit you. Sometimes we will hold the health fair in one place and ask you to come to see us.

THE HEALTH ASSESSMENTS



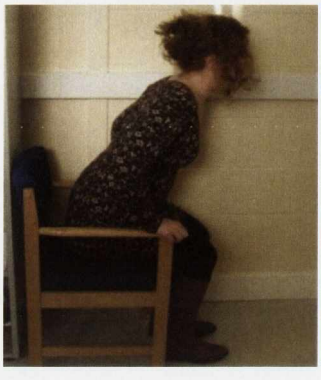
The first assessment is called grip strength. You will be asked to squeeze a device which will measure your strength. This will help us see how strong people are as they get older.



The second assessment will be to measure your waist and hips. This will help us understand the overall health of people.

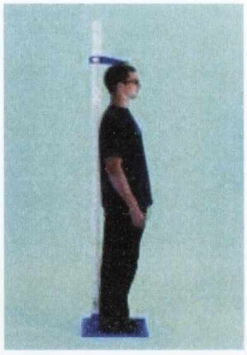


We will then measure your blood pressure.

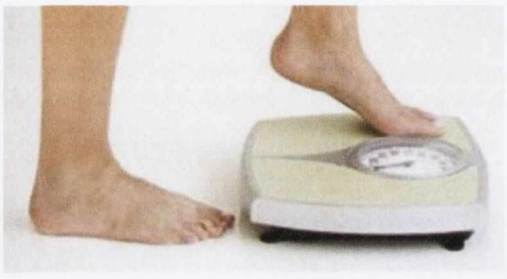


The next assessment is called 'timed get up and go'. This will tell us about your strength and steadiness. This will help us learn if people become frail as they grow older.





We will measure your height. If you cannot stand up we will measure your arm, from this we will be able to tell you your height.



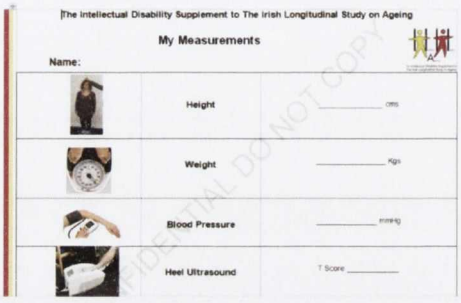
We will then measure your weight. This will help us develop a picture of obesity in the older population.



The final health check is a bone scan of your heel. This will tell us how strong your bones are, which is very important as you grow older.



We will write down what you say, but we will not use your name.



You will get a copy of your results on the day of the assessment.



If you have any questions please do not hesitate to contact me.

Eilish Burke
idstilda@tcd.ie
01-8963187
087-2632241

The Intellectual Disability Supplement to TILDA

The University of Dublin, Trinity College,

School of Nursing & Midwifery,

24 D'Olier Street, Dublin 2.

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The Intellectual Disability Study to TILDA is funded and supported by

Intellectual Disability Supplement to TILDA
The University of Dublin, Trinity College
School of Nursing & Midwifery
24 D'Olier Street, Dublin 2.
Tel: 01 - 8963186/01 - 8963187 Fax: 01 - 896 3001

An Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing

Dear Participant



Thank you for taking part in the Health Fair of Wave 2 of the IDS-TILDA study.



This will help us learn more about people's health as they grow older.

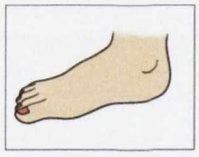
The assessments will take about 30 minutes to complete.



Please read the information booklet enclosed to find out about the assessments



On the day of your appointment we will ask your permission to do the assessments. It is your choice



IMPORTANT
Please do not wear tights as the bone strength assessment will be on your bare foot.



If you have any questions please call me on 087-2632241 or 01 - 8963187.



Yours sincerely,

Eilish

Eilish Burke
Project Manager/PhD student
Intellectual Disability Supplement to TILDA

Health Fair Appointment

IDS-TILDA Health Assessment

An Intellectual Disability Supplement to
The Irish Longitudinal Study on Ageing



Eilish Burke will visit on _____

Date: _____

Time: _____

Venue: _____



If you want some more information please call Eilish on:

087 2632241



Your appointment will take between 20 to 30 minutes.

Thank you very much for your help with this important study

Intellectual Disability Supplement to TILDA
The University of Dublin, Trinity College,
School of Nursing & Midwifery, 24 D'Olier Street, Dublin 2

Telephone: 01-8963186/3187 Fax: 01-8963001

Email: idstilda@tcd.ie

Website : www.idstilda.tcd.ie

Department of Health & Children
AN ROINN SLÁINTE AGUS LEANAÍ

HRB Health Research Board

tilda
The Irish Longitudinal Study on Ageing

Appendix 14:

Pilot Study Comment and Feedback Form

Please tell us what you thought of each of the assessments.

Please tick or mark the smiley face that tells us how you felt about the assessment.

Assessment	I really liked it	It was OK	I did not like it
Grip Strength			
Waist and hip measure.			
Blood Pressure.			
Time up and go.			
Height measure.			
Weight measure.			
Heel scan.			

Please tell us anything else you would like to say about doing the Health Fair.

Appendix 15:

Barthel Index - Modified

Conversion of IDS-TILDA ADLs into a Modified Version of Barthel Index of ADLs scores			
	IDS-TILDA codes	IDS changes to	Modified Barthel Index codes
FL1 Walking 100 yards			**Mobility**
	1=No difficulty	→	3=Independent
	2=Some difficulty	→	2=walks with help
	3=A lot of difficulty	→	1=Wheelchair independent
	4=Cannot do at all	→	0=Immobile
FL11 (want to chat to you about this one- might use earlier variable – several flights of stairs)			**Stairs**
	1=No difficulty	→	2=Independent
	2=Some difficulty	→	1= Needs help
	3=A lot of difficulty	→	0=Unable
	4=Cannot do at all		
FL24			**Dressing**
	1=No difficulty	→	2= Independent
	2=Some difficulty	→	1= Needs help
	3=A lot of difficulty	→	0=Unable
	4=Cannot do at all		
FL 38			**Bathing**
	1=No difficulty	→	1= Independent
	2=Some difficulty		0= Dependent
	3=A lot of difficulty	→	
	4=Cannot do at all		
FL43			**Grooming**
	1=No difficulty	→	1= Independent
	2=Some difficulty		0= Needs help
	3=A lot of difficulty	→	
	4=Cannot do at all		
FL46			**Feeding**
	1=No difficulty	→	2=Independent
	2=Some difficulty	→	1=Needs help
	3=A lot of difficulty	→	0= Dependent
	4=Cannot do at all		
FL51			**Transfer**
	1=No difficulty	→	3= Independent
	2=Some difficulty	→	2=Minor Help
	3=A lot of difficulty	→	1 =Major help
	4=Cannot do at all	→	0=Unable
FL56			**Toileting**
	1=No difficulty	→	2=Independent
	2=Some difficulty	→	1=Needs help
	3=A lot of difficulty	→	0= Dependent
	4=Cannot do at all		
Incontinent- bladder			**Bladder**
	PH90 1=Yes	→	0=Incontinent
	PH91 5=No	→	1=Occasional (In last month)
	PH90 5=No	→	Continent

Incontinent –bowel			Bowel
	PH95 1=Yes	→	0=Incontinent
	PH96 5=No	→	1=Occasional (In last month)
	PH95 5=No	→	Continent

Appendix 16:
Health Fair Certificate of Achievement

INTELLETUAL DISABILITY SUPPLEMENT TO THE IRISH LONGITUDINAL STUDY ON AGEING

THIS CERTIFIES THAT

.....

has successfully completed the Health Assessment
as part of the second Wave of
The Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing
and is therefore awarded this

CERTIFICATE

Eilish Burke

Project Manager/Health Assessor

Prof. Mary McGee

Principal Investigator IDS-TILDA

Appendix 17:
Linear by linear

Linear-by-Linear test of association

	p-value
Gender	**0.033**
Age	**<0.001**
Level of ID	**<0.001**
Type of Residence	**<0.001**
Down Syndrome	**<0.001**
Scoliosis	**0.001**
Parental History of Hip Fracture	0.14
Cerebral Palsy	**0.001**
Epilepsy	**<0.0001**
Asthma	0.528
Stroke &/ TIA	0.326
Hypothyroidism	0.546
Diabetes	**0.017**
Menopause	0.661
All Cancer	0.291
Difficulty walking 100 yards	**<0.001**
Mobility Aid/Wheelchair user	**<0.001**
Antiepileptic Medications	**<0.001**
Ever smoked	0.454
Dairy regulations	0.406
Gastro reflux	**0.025**
Stomach ulcers	0.336
BMI	**<0.001**
waist classification	0.209
Activity level	**<0.001**
Grip strength	**0.053**
Proton pump	**<0.001**

The linear-by-linear association test has more statistical power than the Pearson chi-square test for the evaluation of ordered variables against alternatives of monotone trend but the more commonly used approach, Pearson's chi-squared, is presented in this thesis. Results are qualitatively unchanged.