

Oral hygiene interventions for people with disabilities: A Scoping Review, a Cochrane Review and a Realist Review

Volume I: Thesis

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

January 2019

Catherine Waldron
RDH, MSc. MA (Health Promotion)

School of Dentistry,
Faculty of Health Sciences,
University of Dublin,
Trinity College.

Supervisors:
Dr. Caoimhin Mac Giolla Phadraig,
Prof. Catherine Comiskey, Prof. June Nunn

Declaration

I declare that this thesis has not been submitted as an exercise for a degree at this or any other university. The work described in this thesis, except where duly acknowledged, is my own.

I agree to deposit this thesis in the University's open access institutional repository or allow the library to do so on my behalf, subject to Irish Copyright Legislation and Trinity College Library conditions of use and acknowledgement.

I consent to the examiner retaining a copy of the thesis beyond the examining period, should they so wish (EU GDPR May 2018).

Catherine Waldron

Date

Summary

Introduction: The starting point for this thesis was a lack of evidence in relation to many aspects of oral health interventions for people with disabilities. Thus, the purpose of the thesis was, firstly, to identify the existing literature in this previously un-researched area (a Scoping Review), secondly, to determine the effect of oral hygiene interventions for a sub-group of people with disabilities, those with intellectual disabilities (ID) (a Cochrane Review) and, finally, to develop a contextual understanding of the mechanisms that may explain the different outcomes in some of the similar interventions in different settings - carer-led oral hygiene interventions for those with ID - by developing theories about why, in what circumstances and for whom, the interventions might work (a Realist Review).

The Scoping Review: The main purpose of a Scoping Review is to quantify the elements of the existing studies on a broad research topic, the search terms are broad and it includes all study designs. The focus of the Scoping Review in this thesis was oral health interventions for children and adolescents with disabilities.

The review revealed a focus on therapeutic interventions for populations with disabilities (46%), however due to the diversity of the therapeutic interventions, relatively small numbers of studies devoted to each type, with a specific disability population, were identified. Skill-based interventions (28%), of which most were focused on oral hygiene skills or methods to teach these skills (90%), and most for a population with ID (48%), determined the focus of the Cochrane Review in this thesis.

The Cochrane Review: A Cochrane Review has a specific research question, with pre-determined inclusion criteria. It gathers all the available evidence, following a systematic approach, thus reducing bias, and results in findings that are more reliable than the individual studies on their own.

The objective of the Cochrane Review completed for this thesis was to assess the effects of oral hygiene interventions for people with ID. While inclusion of a wide range of study designs has resulted in the findings being reported in different formats (narrative and meta-analyses), it has resulted in a very comprehensive review of the outcomes of oral hygiene interventions for people with ID.

The 34 studies identified, showed: some benefits when a carer of a person with ID used a special manual toothbrush; inconsistent findings between outcomes when an electric toothbrush was used by or for people with ID; carers oral health knowledge consistently improved following training. However, this training did not always impact on the carers' attitude or behaviour and resulted in only small reductions in the levels of gingival inflammation of the people with ID for whom they cared, the benefit to the

people with ID was shown to be greater when some level of monitoring of the carer's behaviour was undertaken.

Of the six studies assessing the effects of teaching people with ID to brush their teeth, the Review confirmed that people with ID can acquire the skills to undertake some or all of the steps involved in toothbrushing.

The Realist Review: The purpose of a Realist Review is to consider how, why, when and for whom comparable interventions work, rather than if they work. It does this by looking closely at the contexts and mechanisms within the interventions, to see if there are any patterns that might impact on the outcomes. These patterns allow theories to be developed, which can then be tested in future interventions.

Based on the findings in relation to the role played by carers in the Scoping and Cochrane reviews, the interventions of interest in the Realist Review carried out for this thesis were carer-led oral hygiene interventions for people with ID.

The review provided a valuable insight into the contexts and mechanisms that affect the carers attitude and behaviour in relation to providing consistent support, assistance or direct care to people with disabilities, the physical and emotional toll on the carers, as well as the ethical dilemmas of providing this consistent level of care to people with disabilities, who may often not understand the need and thus resist the care.

The theories developed from this review could additionally, be applied to interventions involving the carers of populations with long-term health conditions such as elderly people, people with dementia and people with physical conditions limiting their level of self-care.

Conclusion: The three reviews together have provided a comprehensive insight, which did not exist previously, into oral hygiene interventions for people with ID: the types of interventions and populations targeted; the clinical and behavioural outcomes; and some contexts and mechanisms that have emerged as crucial to ensure success in future interventions. Each review has provided information that would not have been discovered by the others alone.

Reviewing evidence using parallel approaches has provided depth to the findings reported in this thesis. All of the review approaches have their strengths and limitations: using them in combination, I believe, has resulted in a whole that is greater than the sum of their parts.

This thesis provides evidence to guide both practical and theoretical approaches for future oral hygiene interventions for people with intellectual disabilities.

Dedication

To my siblings, Rebecca, Mary and Mark, who regularly appeared from near and far, to cook gourmet meals, stock up my cupboards and generally keep my life on track when I could not!

To my dear friends, Anna, Anne, Úna and Kevin, who were always there to cheer me up.

To my supervisors, Kev, June and Catherine whose inspiration, patience and encouragement never waned.

Thank you one and all for your support;
I could not have done it without you.

Acknowledgments

No words can fully describe the gratitude I feel towards Prof. June Nunn and Dr. Caoimhin Mac Giolla Phadraig, who not only supervised my PhD but also were review team members on all three reviews, and to my third supervisor, Prof. Catherine Comiskey who was also a team member on both the Scoping and Cochrane Reviews. Their constant support and encouragement, the massive input of their time and generous sharing of their expertise is gratefully acknowledged.

I am indebted to all the Cochrane Review team members, each brought specific expertise and insight to the review and gave their time generously to the process: Prof. June Nunn, Dr. Caoimhin Mac Giolla Phadraig, Prof. Catherine Comiskey, Prof. Mike Clarke, Dr. Maria van Harten, Dr. Suzanne Guerin and Dr. Erica Donnelly-Swift.

I acknowledge the assistance of The Cochrane Oral Health Group in the production of the Cochrane protocol and review, particularly Laura MacDonald, Anne Littlewood, Professor Helen Worthington, Professor Jan Clarkson, Dr Tanya Walsh and Dr Martin McCabe. Also, those who assisted by translating the articles written in languages other than English.

I appreciate the time and effort made by authors of included and excluded studies for the Cochrane Review to source data and provide additional information.

I am very grateful to the oral healthcare professionals who gave so willingly of their time and expertise during the Realist Review - The Expert Panel: Catherine Binkley (USA), Conac Bradley (Ireland), Dominique Declerck (Belgium), Imke Kaschke (Germany), Lorna McPherson (Scotland), Johanna Norderyd (Sweden), Archana Pradhan (Australia), Shelagh Thompson (England) and to Siobhan Stapleton (Ireland) who piloted the Expert Panel survey. The Local Experts: Triona McAllister, Sandra Coen, Colette Long, Danielle McGeown, Antoinette Nolan, Mary Ormsby and Gillian Smith.

I acknowledge the considerable assistance of the library staff in the Dublin Dental University Hospital in sourcing articles from our own library, Trinity College Library and via inter library loans - Isabel Fleischmann, Caroline Meehan, Caroline Montgomery, Fleur Finlay and Sinead Magee.

I will be forever grateful to the Dublin Dental University Hospital and Trinity College for awarding me the 1252 Student Scholarship which allowed me to undertake this PhD.

Content

Declaration	i
Summary	ii
Dedication	iv
Acknowledgments	v
List of Tables	xi
List of Figures.....	xiv
Chapter 1 Introduction	1
1.1 Introduction.....	1
1.2 Background.....	2
1.2.1 Definition of Disability.....	2
1.2.2 Definition of Intellectual Disability (ID)	4
1.2.3 Prevalence of Disabilities.....	5
1.2.4 The role of carers for those with disabilities.....	6
1.3 The role of oral hygiene in general and oral health	7
1.3.1 Assessment of oral health	8
1.4 The oral health of people with disabilities.....	9
1.4.1 Oral health of children with disabilities.....	9
1.4.2 Oral health of children with intellectual disabilities.....	11
1.4.3 Oral health of adults with intellectual disabilities	12
1.5 Oral health interventions	15
1.5.1 Oral Health Interventions and Disability	15
1.5.2 Evaluation of oral health interventions	16
1.6 Health behavioural change.....	17
1.6.1 Behaviour change theories	18

1.6.2	Behaviour change and people with ID	19
1.7	Rationale for this research	20
Chapter 2 – General Methodology	22	
2.1	Introduction.....	22
2.2	Choosing multiple review methodologies.....	24
2.3	A Scoping Review.....	25
2.3.1	The focus of the Scoping Review	26
2.4	A Cochrane Review	27
2.4.1	The focus of the Cochrane Review:	28
2.5	A Realist Review	29
2.5.1	Why a realist approach was undertaken.....	31
2.5.2	The focus of the Realist Review.....	34
Chapter 3 A Scoping Review	36	
3.1	Introduction.....	36
3.2	Methods	36
3.2.1	Inclusion and Exclusion Criteria	36
3.3	Study Selection	38
3.4	Screening.....	39
3.5	Data Extraction	41
3.6	Results.....	42
3.6.1	Participants	42
3.6.2	Interventions.....	43
3.6.3	Outcomes	44
3.6.4	Timeline	45
3.7	Discussion.....	45
3.7.1	Limitations	46
3.8	Conclusions.....	47
Chapter 4 Cochrane Review.....	49	

4.1	Introduction.....	49
4.2	Background.....	50
4.2.1	Description of the condition.....	50
4.2.2	Definition of intellectual disability.....	51
4.2.3	Barriers to oral hygiene practice for people with ID	53
4.2.4	Facilitators for oral hygiene practice for people with ID	53
4.3	Description of the intervention	54
4.3.1	How the intervention might work	54
4.4	Why it is important to do this review	56
4.5	Methods	57
4.5.1	Criteria for considering studies for this review	57
4.5.2	Search methods for identification of studies	61
4.5.3	Data collection and analysis	62
4.6	Results.....	69
4.6.1	Description of studies	69
4.6.2	Results of the search.....	69
4.6.3	Included studies	71
4.6.4	Excluded studies.....	86
4.6.5	Risk of bias in included studies	87
4.7	Effects of interventions.....	94
	Comparison 1: Special manual toothbrushes versus conventional manual toothbrushes for people with ID	96
	Comparison 2: Electric toothbrushes versus manual toothbrushes for people with ID	113
	Comparison 3: Oral hygiene care training versus no oral hygiene care training or a placebo training session for carers of people with ID.....	131
	Comparison 4: Oral hygiene training versus no oral hygiene training for people with ID	148
	Comparison 5: One-, three- and six-monthly dental recall intervals versus no dental recall for people with ID	156
	Comparison 6: Use of individual clinical photographs as a toothbrushing motivator versus no motivator for people with ID	165

Comparison 7: Daily, twice weekly and once weekly brushing frequency versus usual care for people with ID	165
Comparison 8: Use of a toothpaste with a plaque disclosing agent versus a conventional toothpaste for people with ID.....	169
Comparison 9: Individualised oral care plan versus usual care for people with ID	169
4.8 Summary of main results	172
4.9 Discussion.....	175
4.9.1 Overall completeness and applicability of evidence.....	176
4.9.2 Validity of assessments used	178
4.9.3 COM-B Characteristics identified in the interventions	178
4.9.4 Quality of the evidence	180
4.9.5 Potential biases in the review process.....	181
4.9.6 Agreements and disagreements with other studies or reviews	182
4.10 Conclusion	185
Chapter 5 A Realist Review	189
5.1 Introduction.....	189
5.1.1. The aim of the Realist Review.....	190
5.2 Methods	190
5.3 Step 1. Identifying Candidate Theories	192
5.4 Step 2: Searching for data	212
5.5 Step 3: Data Management.....	215
5.6 Step 4: Data Synthesis	219
1. Organisational involvement	223
2. Policy and guidelines	227
3. Make it meaningful.....	228
4. Needs of people with ID.....	232
5. Caring for carers	234
6. A pat on the back.....	238
7. Achieving goals.....	241

8. Support from whom?	244
9. Remind them.....	247
10. Everyone's voice	251
5.7 Step 5: Refined Theories	253
5.7.1 Summary of the Mechanisms, Contexts and Outcomes	253
5.7.2 Candidate and Refined Theories following synthesis	255
5.7.3 Framework for Refined Theories.....	259
5.8 Discussion.....	262
5.9 Conclusions.....	263
 Chapter 6 Discussion and Conclusions.....	 266
6.1 Introduction.....	266
6.2 Individual Review Discussion.....	267
6.2.1 The Scoping Review	267
6.2.2 The Cochrane Review	268
6.2.3 The Realist Review	270
6.3 Overall Discussion.....	272
6.3.1 The involvement of carers.....	273
6.3.2 Stakeholder involvement	273
6.3.3 Outcomes measured	274
6.3.4 Limitations	275
6.3.5 Additions to existing knowledge	276
6.3.6 Recommendations.....	280
6.4 Conclusions.....	281
 Chapter 7 References	 281
7.1 General References	281
7.2 Scoping Review: Included Studies	293
7.3 Cochrane Review: Included Studies	300
7.4 Realist Review: Included Studies	302

List of Tables

Table 1: Description of intellectual disability as per ICD-10.....	4
Table 2: Irish and worldwide dental caries experience by age groups.....	10
Table 3: Sequelae of dental caries in Ireland.....	13
Table 4: Gingival disease in ID and general adult populations	14
Table 5: Steps in a Scoping Review (Arksey & O'Malley 2005).....	26
Table 6: Screening protocol	38
Table 7: Scoping Review search terms used in the PubMed database	39
Table 8: Types of disabilities identified in the interventions.....	42
Table 9: Groups other than CAWD involved in the interventions	43
Table 10: Types of interventions	44
Table 11: Outcomes measured.....	44
Table 12: Stages and time frames for the Cochrane Review.....	49
Table 13: The ICD-10 levels of ID.....	52
Table 14: Modifications to the Gingival Inflammation indices used	79
Table 15: Modifications to the Plaque indices used	81
Table 16: Summary table of Comparisons	94
Table 17: C1 Gingival inflammation studies - short term	99
Table 18: C1 Gingival inflammation studies - medium term	101
Table 19: C1 Plaque studies - short term.....	103
Table 20: C1 Plaque studies - medium term.....	105
Table 21: C1 Plaque NRS - medium term.....	105
Table 22: C1 Oral health assessment studies	107
Table 23: C1 Behaviour studies - medium term.....	109
Table 24: Comparison 1 – All outcomes/ subgroups effect estimates.....	112
Table 25: C2 Gingival inflammation studies - short term	116
Table 26: C2 Gingival inflammation studies - medium term	117
Table 27: C2 NRS Gingival inflammation studies - < 12 weeks.....	119
Table 28: C2 NRS Gingival inflammation studies - >12 weeks.....	120
Table 29: C2 Plaque studies - short term.....	122
Table 30: C2 Plaque studies - medium term.....	123
Table 31: C2 NRS Plaque studies - < 12 weeks	125
Table 32: C2 NRS Plaque studies - > 12 weeks	126
Table 33: C2 Calculus studies - medium term	128

Table 34: C2 All outcomes and subgroups effect estimates.....	130
Table 35: C3 Gingival inflammation – medium term	134
Table 36: C3 Plaque – medium term	136
Table 37: C3 NRS Plaque – short term.....	138
Table 38: C3 NRS Plaque – medium term.....	140
Table 39: C3 Knowledge – short and medium term	141
Table 40: C3 Behaviour – short term	144
Table 41: C3 Behaviour, attitude and self-efficacy – medium term	144
Table 42: C3 NRS Behaviour and self-efficacy – medium term	146
Table 43: C3 All outcomes and subgroups effect estimates.....	147
Table 44: C4 Gingival inflammation – short term	149
Table 45: C4 Plaque – short term.....	151
Table 46: C4 NRS Self-efficacy – medium and long term.....	153
Table 47: C4 All outcomes with effect estimates	155
Table 48: C5 Gingival inflammation – long term.....	158
Table 49: C5 Gingival bleeding – long term	158
Table 50: C5 Plaque – long term.....	161
Table 51: C5 Calculus – long term.....	161
Table 52: C5 All outcomes and subgroup effect estimates	164
Table 53: C6 Plaque – medium term	166
Table 54: C6 All outcomes and subgroup effect estimates	166
Table 55: C7 Gingival inflammation – short term	168
Table 56: C8 Gingival inflammation – short term	170
Table 57: C8 Plaque – short term.....	170
Table 58: C9 Plaque – medium term	171
Table 59: First Draft of Candidate Theories	196
Table 60: Revised Draft of Candidate Theories.....	199
Table 61: Expert Panel members	209
Table 62: The search terms used to filter the Cochrane Review findings....	213
Table 63: Realist Review search terms.....	214
Table 64: Realist Review studies & input into the Candidate Theories	221
Table 65: Behaviour change techniques relevant to Theory 3	230
Table 66: Behaviour change techniques relevant to Theory 6	240
Table 67: Behaviour change techniques relevant to Theory 7	243
Table 68: Behaviour change techniques relevant to Theory 9	249

Table 69: Mechanisms at the individual carer level and linked theories.....	253
Table 70: Mechanisms at the Organisational level and linked theories	254
Table 71: Context for Organisations and Carers	255
Table 72: Outcomes reported/referred to in the studies.....	255
Table 73: Candidate and Refined Theories based on synthesis	256
Table 74: Findings compared to other systematic reviews.....	278

List of Figures

Figure 1: Prisma Flow Chart for the Scoping Review	41
Figure 2: Number of interventions over the decades	45
Figure 3: Prisma Flow Chart for the Cochrane Review	70
Figure 4: Cochrane Review Overall Risk of Bias.....	88
Figure 5: Cochrane Review Individual Studies Risk of Bias	93
Figure 6: C1 Meta-analysis Gingival inflammation - short term	100
Figure 7: C1 Meta-analysis Gingival inflammation - medium term	102
Figure 8: C1 Meta-analysis Plaque - short term.....	104
Figure 9: C1 Meta-analysis Plaque - medium term	106
Figure 10: C1 Meta-analysis Oral health assessments.....	108
Figure 11: C1 Meta-analysis Behaviour – carer elements	110
Figure 12: C1 Meta-analysis Degree of Resistance of people with ID.....	111
Figure 13: C2 Meta-analysis Gingival inflammation - medium term	118
Figure 14: C2 Meta-analysis NRS Gingival inflammation - long term.....	121
Figure 15: C2 Meta-analysis Plaque - medium term.....	124
Figure 16: C2 Meta-analysis NRS Plaque - medium term.....	127
Figure 17: C2 Meta-analysis Calculus - medium term	129
Figure 18: C3 Meta-analysis Gingival inflammation – medium term.....	135
Figure 19: C3 Meta-analysis Plaque – medium term	137
Figure 20: C3 Meta-analysis NRS Plaque – short term	139
Figure 21: C3 Meta-analysis Knowledge – short term	142
Figure 22: C3 Meta-analysis Knowledge – medium term	143
Figure 23: C3 Meta-analysis Behaviour, attitude and self-efficacy – medium term.....	145
Figure 24: C4 Meta-analysis Gingival inflammation – short term	150
Figure 25: C4 Meta-analysis Plaque – short term	152
Figure 26: C4 Meta-analysis NRS Self-efficacy (time frame unclear)	154
Figure 27: C5 Meta-analysis Gingival inflammation – long term	159
Figure 28: C5 Meta-analysis Gingival bleeding – long term.....	160
Figure 29: C5 Meta-analysis Plaque – long term.....	162
Figure 30: C5 Meta-analysis Calculus – long term	163
Figure 31: C6 Meta-analysis Plaque and Extrinsic stain – medium term.....	167
Figure 32: Realist Review steps	191

Figure 33: Intervention Change Chain (McMahon & Ward 2012).....	197
Figure 34: Realist theory refining process diagram (Wong <i>et al</i> 2013).....	212
Figure 35: Prisma Flow Chart for the Realist Review	215
Figure 36: Framework for Theories with key Contexts and Mechanisms....	261

Chapter 1 Introduction

1.1 Introduction

Worldwide, the oral health of people with disabilities is poorer than the general population (Crowley *et al* 2005, De Jongh *et al* 2008, Hennequin *et al* 2008, Anders & Davis 2010, Morgan *et al* 2012, Oliveira *et al* 2013, Campanaro *et al* 2014). An increasing number of oral health interventions, specifically designed to improve the oral health of people with disabilities, have been implemented and evaluated in the last two decades. However, the effectiveness of these oral health promotional, educational and therapeutic interventions has not been systematically examined.

The complex nature of oral health interventions designed to prevent dental diseases, for the general population, which require individuals to make behavioural changes, learn skills, continue to apply these over long periods of time and allow for other external influences, makes it difficult to identify and assess the outcomes of relevance. Interventions have been shown to be successful in one setting and not another, or to have different outcomes when delivered by different teams or personnel (Watt & Marinho 2005).

The design of preventive oral health interventions can be even more challenging for people with disabilities, when access to care is more difficult, the interventions are often applied to one person (the carer) with the intention of changing the health of another (the person with disabilities), time and resources are stretched for parents, carers and healthcare professionals alike, the consequences of oral diseases are more profound and an awareness or understanding of the oral health problems that may arise, on the part of people with disabilities and their carers, is lacking (Kaye *et al* 2005, Dougall & Fiske 2008a, Thole *et al* 2010, Forsell *et al* 2011, Prabhu *et al* 2010, Sagheri *et al* 2013).

This makes it especially important that these interventions are reviewed systematically. Three different systematic review methods were used in this

review study in an effort to fully understand the research topic; both quantitative and qualitative data were explored.

The purpose of this review study was firstly, to identify the existing literature in this previously un-researched area (a Scoping Review), secondly, to determine the effect of oral hygiene interventions for a sub group of people with disabilities, those with intellectual disabilities (ID) (a Cochrane Review) and, finally, to develop a contextual understanding of the mechanisms that may explain the different outcomes found for similar interventions in different settings, by developing theories about why, in what circumstances and for whom, the interventions might work, for a more focused group of interventions; carer-led oral hygiene interventions for those with ID (a Realist Review).

It is hoped that the findings of these reviews will assist those designing, implementing and funding future oral health interventions and those caring for people with disabilities, with the potential to ensure more positive and sustained outcomes for the oral health of this subset of the general population.

1.2 Background

The purpose of this chapter is to provide background information important to the research topic. It will define the key elements, outline the existing knowledge in relation to the oral health status of people with disabilities and discuss the complexities and challenges that may be encountered when planning, implementing or evaluating oral health interventions for this population.

1.2.1 Definition of Disability

The World Health Organisation (WHO) stated in their World Report on Disability (2011) that there was no agreement on the definition of disability, despite the enormity of the issue, making comparisons between studies on the topic difficult.

There has been a gradual shift from considering disability under a medical model, to a social model and ultimately to a biopsychosocial model. The current WHO definition of disability, introduced in 2001, is the International Classification of Functioning, Disability and Health (ICF), which incorporates the complex interactions between health conditions, environmental factors and personal factors that combine to define an individual. The ICF covers a number of domains: dysfunction at one or more of the following levels: the body or body part (impairments), the whole person (activity limitations), or the whole person in a social context (participation restrictions) (WHO 2001).

At its introduction, the ICF was seen as a 'workable compromise' between the medical and social models and is promoted as a 'bio-psycho-social model' to conceptualise impairment and disability. The ICF conceptual framework defines disability as:

The negative aspects of the interaction between an individual (with a health condition) and that individual's contextual factors (personal and environmental factors). Impairments are interactions affecting the body; activity limitations are interactions affecting individual's actions or behaviour; participation restrictions are interactions affecting person's experience of life.

This biopsychosocial model, although more holistic, can be difficult to apply to research populations as the elements described are very broad and can be difficult to identify or measure. The data collected in studies may be influenced by the purpose of the research or the way the questions are posed; disability is not a dichotomous condition.

The WHO has also developed an operational definition of disabilities: The International Statistical Classification of Diseases and Related Health Problems, now in its 10th version (ICD10) (WHO 2011). This is an exhaustive list of human diseases. The ICF definition of disability is designed to complement the ICD-10 definition.

Applying the ICF model to research populations can be complex and implementing it has been slow. Additionally, the concept is relatively new, thus any review of past literature, on the prevalence of disabilities or the effectiveness of interventions for those with disabilities, is unlikely to have used the ICF framework to any great extent. Discussion on how to incorporate these contextual factors into data collection for this population continues with the development of core outcome sets (Faulks *et al* 2013, Vale *et al* 2015). Until such time as these constructs are in general use, in order to determine the inclusion or exclusion of studies in the reviews undertaken in this thesis, it was decided therefore to use the ICD-10 definition of disability as the benchmark, since the ICD-10 provides a detailed nomenclature and classification of conditions (Kurbasic *et al* 2008).

1.2.2 Definition of Intellectual Disability (ID)

In relation to defining ID specifically, three elements are common: a significant impairment of intelligence, a resultant significant reduction in adaptive behaviour/social functioning and the development of the condition before the age of 18, which persists throughout life. The ICD-10 describes four levels of ID (Table 1) (WHO 1992).

Table 1: Description of intellectual disability as per ICD-10

Mild ID (ICD 10 F70): IQ range 50-69	Ability to use speech in everyday situations; usually full independence in self-care; Difficulties in identification of this population arise, as those with borderline disability may not be in contact with service providers.
Moderate ID (ICD 10 F71): IQ between 35-49	Slow in comprehension; supervision of self-care, retarded motor skills.
Severe ID (ICD 10 F72): IQ between 20-34	Marked impairment of motor skills; clinically significant damage to Central Nervous System (CNS).
Profound ID (ICD F73): IQ less than 20	Severely limited understanding; Immobility or restricted mobility; Incontinence; requires constant supervision; usually organic aetiology.

1.2.3 Prevalence of Disabilities

The prevalence of disabilities can be difficult to accurately assess: results can vary depending on the type and range of questions asked in any survey. For example, comparing the results of the Census collected in Ireland in 2006 and the data from the subsequent National Disability Survey (NDS) (Central Statistics Office 2006, National Disability Survey 2008); the 2006 census included two questions on disability relating to the presence of a long-term health condition and the impact of that condition on functioning; the NDS defined disability as difficulties in functioning in any of the following nine domains:

1. Seeing
2. Hearing
3. Speech
4. Mobility and dexterity
5. Remembering and concentrating
6. Intellectual and learning
7. Emotional, psychological, and mental health
8. Pain
9. Breathing.

The disability prevalence rate of 9.6% in the Census rose to 18.5% when the NDS figures were extrapolated to the general population (CSO 2006, NDS 2008). Children with disabilities were found to be under-represented in the census. The prevalence figure of 18.5% is more in line with disability levels quoted in other countries (NDS 2008). The definition of disability in the more recent 2016 Irish census was revised; disabilities were grouped under seven 'long-lasting conditions or difficulties' and respondents were asked how these disabilities affected them in relation to four activities of daily living. The 2016 Census reported that 13.5% of the population indicated that they had a disability (CSO 2016). The National Intellectual Disability Database (NIDD) in Ireland showed there were 28,275 people registered on the database at the end of December 2016. Based on 2011 Census of Population figures, this represents a prevalence rate of 6.16 per 1,000 population (Health Research Board 2017).

The ability of national surveys to more accurately represent the prevalence of disabilities worldwide has improved. These surveys show that the number of people with disabilities is increasing (WHO 2011). This, it is suggested, is due to the increasing life span of those with disabilities. These more accurate data should inform decision making in relation to the provision of care for this population group. Assessing the quality and outcomes of existing interventions targeted at the health of people with disabilities is therefore important, to ensure that the resources are directed at the most effective care programmes.

1.2.4 The role of carers for those with disabilities

Disability by its nature implies difficulty in engaging in certain activities of daily living. In the context of this study, oral care is one such activity. People with disabilities may have more difficulty completing the oral hygiene routines required on a daily basis, which are essential to maintaining oral health and preventing oral diseases. They may require the assistance of others to perform this routine (Crowley *et al* 2005).

Those caring for people with disabilities can be grouped into informal or formal carers. An informal carer may be defined as a person who is providing unpaid care, including personal care, to a person who is in need of that care in the home due to illness, disability or frailty. Other terms used for informal carers in the literature are 'unpaid carer', 'family carer', 'lay carer' or simply 'carer'. Informal carers may assist others in the activities of daily living (ADL) such as feeding, toileting, dressing, grooming, maintaining continence, showering or bathing, walking and transferring from bed to chair or wheelchair. They may also assist with the more complex instrumental activities of daily living (iADL) such as managing finances, shopping, preparing meals, housekeeping, taking medications and arranging transport and activities. Health strategists increasingly acknowledge the contribution of informal carers in the daily lives of people with disabilities.

The exact definition of informal carers used in surveys of the number of people providing unpaid care in populations varies from jurisdiction to jurisdiction, resulting in figures ranging between 4.1% (Ireland, 2011), 15.5% (Europe,

2007) and 28.5% (USA, 2009), (Scottish Government 2010, Care Alliance Ireland, 2013, Family Caregiver Alliance, USA 2016).

A formal carer is more likely to be associated with a system service, such as social or health services, or a private company, caring for a person either in their home or in another setting, most often receiving payment, but such services may occasionally also include volunteers. Their role is similar to the informal carer but they may have received some qualification or training to carry out this role, which can be regulated. Other terms used for formal carers in the literature are 'health care workers' or 'paid carers' (Scottish Government 2010, Care Alliance Ireland, 2013, Family Caregiver Alliance, USA 2016).

Carers, whether informal or formal, may not have the skills or motivation required to provide oral hygiene assistance or carry out oral hygiene routines for people with disabilities (Cumella *et al* 2000, Pradhan *et al* 2009, Thole *et al* 2010, Minihan *et al* 2014, Zaihan *et al* 2015).

1.3 The role of oral hygiene in general and oral health

The two most common oral diseases are dental caries (tooth decay) and periodontal disease (gum disease), both of which are preventable (Petersen 2003). Dental caries is usually first experienced at a young age, while periodontal disease is considered more a disease of adulthood. Dental plaque has been shown to have a major role in the development of both periodontal disease and dental caries (Löe *et al* 1965, Löe 2000, Axelsson *et al* 2004, Broadbent *et al* 2011, Van Der Weijden & Slot 2011).

Oral hygiene is the process of cleaning the hard and soft tissues of the oral cavity (teeth, gums, and tongue) as well as any fixed dental prostheses, oral appliances and dentures. Failure to perform regular and effective oral hygiene will result in increased levels and varieties of the dental organisms that make up dental plaque. The definition of oral hygiene used in this research is the mechanical removal of plaque with either manual or electric toothbrushes, interdental aids or other mechanical aids. The impact of chemical agents on oral hygiene will be discussed in chapter 4.

Poor oral hygiene may have an effect on a person's quality of life due to pain and discomfort when eating, poor self-esteem or sleep disturbance as a consequence of pain from dental caries (Bonetti *et al* 2015). There is also evidence linking levels of oral hygiene and periodontal status to diabetes, aspiration pneumonia and rheumatoid arthritis, as well as emerging evidence for a link with other systemic diseases and conditions, such as cardiovascular disease, liver disease and obesity (Horwitz *et al* 2000, Borgnakke & Wenche 2015).

In the general population, brushing effectively and regularly with fluoridated toothpaste has been shown to play an important role in preventing dental caries and periodontal disease (Löe *et al* 1965, Chestnutt *et al* 1998, Löe 2000, Axelsson *et al* 2004, Broadbent *et al* 2011, Van Der Weijden & Slot 2011, Zimmermann *et al* 2015). This health-related behaviour requires an individual to have the relevant knowledge, motivation and skills to perform the necessary routines (Stewart *et al* 1996, Watt & Marinho 2005, Broadbent *et al* 2011). The challenge in relation to preventing dental caries and periodontal disease is even greater for people with disabilities when all these elements are considered.

1.3.1 Assessment of oral health

The following is a brief overview of the elements of dental caries and periodontal disease most commonly used to assess oral health as reported in oral health interventions.

Dental Caries

The most commonly used epidemiological index to assess dental caries experience in populations is the Decayed, Missing and Filled index (DFMT/dmft), which is a count of the number of permanent/primary teeth that are either decayed, missing or filled as a consequence of dental caries. The oral examination may include the use of a dental probe and air drying of the tooth or be limited to a visual examination, which is indicated by a number and subscript in the quoted index (D3vcMFT). The index expresses a person or populations' dental caries experience over a lifetime. It provides information in relation to the prevalence of disease, treated and untreated, and may assist

in the determination of future resources required for its management, as well as the impact of interventions.

Periodontal Disease

There are many indices of periodontal health used in the literature: these include specific assessments of plaque, gingival inflammation, gingival bleeding, loss of attachment, calculus and bacterial concentrations. The indices may assess all teeth fully erupted in the mouth or may assess only a selected number of teeth, most commonly the six 'standard teeth'. These standard teeth are regarded as representing the whole mouth and have been validated in a number of studies. The indices may also assess a variety of tooth surfaces (Wei & Lang 1981, Wei & Lang 1982). The complexity of assessments used in oral health research studies can make it difficult to compare findings between studies. When reporting the findings of the interventions in this thesis, every effort will be made to clearly report the number of teeth or tooth surfaces assessed as well as the index and scale used.

1.4 The oral health of people with disabilities

1.4.1 Oral health of children with disabilities

In Ireland, no national data exist in relation to the oral health of preschool children. However, in relation to the oral health of pre-school children with disabilities in Ireland specifically, a few small-area oral health surveys have been conducted. Comparing the data for the two more recent of these studies; one found that dental caries was not detected in children with disabilities before the age of four, however, at the age of 5 and 7 years, 25% and 37% of children with disabilities, respectively, had experienced dental caries (Sagheri *et al* 2013). The other study recorded a caries experience of between 12% to 23%, with a mean dvmft of 0.06 (SD 0.31) for 3-year-olds and a mean dmft of 0.52 (SD 1.27) for 5-year-olds (Stapleton 2015).

Looking at the data for school-aged children, Whelton *et al* (2009) reported in an Irish study, carried out in 2003, that 32% of 5-year-olds with profound ID attending special schools or day care centres, and 27% of 5-year-olds with moderate ID, had experienced dental caries. By comparison, data from the

2002 survey of general child oral health in Ireland indicated an even higher prevalence of dental caries in 5-year-olds in fluoridated and non -fluoridated areas of 37% and 51%, respectively. (Whelton *et al*, 2006).

The finding of a lower prevalence of dental caries in Irish preschool and young children with disabilities, compared to their counterparts in the general population, continues into the older age groups of children and adolescents: 49% and 58% of 12 and 15-years-olds with disabilities, respectively, compared to 54% and 75% of 12 and 15-year-olds respectively, in the general population, are reported as having experienced dental caries (Whelton *et al* 2006, Whelton *et al* 2009).

When comparing these Irish data to that from other countries, for the most part the data show that there is a similar, lower prevalence of dental caries experience in populations with disabilities, compared to similar age groups in general populations (McAlister & Bradley 2003, Dye *et al* 2004, Whelton *et al* 2006, Parnell *et al* 2007, De Jongh *et al* 2008, Hennequin *et al* 2008, Sagheri *et al* 2013, Public Health England 2015, Stapleton *et al* 2015, Marks *et al* 2018), (Table 2).

Table 2: Irish and worldwide dental caries experience by age groups

Published Study and Year	Country	Year data collected	5-year-olds with dental caries	12-year-olds with dental caries	15-year-olds with dental caries
Grey bars indicate general population groups (without disabilities)					
McAlister & Bradley 2003	Ireland	2001	32%	49%	61%
Whelton <i>et al</i> 2009	Ireland	2003	27% – 32%	49%	58%
Parnell <i>et al</i> 2007	Ireland	2002	41% – 51%	-	-
Whelton <i>et al</i> 2006	Ireland	2002	37% -51%	54%	75%
Dye <i>et al</i> 2007	USA	2004	28% 2-5-yr-olds	51% 6-11-yr-olds	-
De Jongh <i>et al</i> 2008	Holland	2004	-	70%	-

				4-12-yr-olds	
Hennequin <i>et al</i> 2008	France	2005	-	18% 6-12-yr-olds	24% 13-20-year-olds
Hennequin <i>et al</i> 2008	France	2005	-	13% 6-12-yr-olds	-
Sagheri <i>et al</i> 2013	Ireland	2006	25%	-	-
Public Health England 2015	England	2013	31%	34%	46%
Stapleton <i>et al</i> 2015	Ireland	2013	12%- 23%	-	-
Marks <i>et al</i> 2017 (Untreated decay)	World	2007-2015	-	21%-67%	25% - 61%

In relation to gingival health, McAlister & Bradley (2003) reported that children with disabilities had high plaque and CPITN scores, 91% of 12-year-olds and 95% of 15-year-olds were assessed as having some level of gingival disease. Whelton *et al* (2009) similarly reported poor levels of oral hygiene with 82% of 12-year-olds and 87% of 15-year-olds requiring some level of treatment ranging from oral hygiene instruction to urgent care. This compares to 71% and 66% for the same age groups in the general population.

1.4.2 Oral health of children with intellectual disabilities

Many of the research studies on the oral health of children with disabilities are based predominantly on populations with intellectual disabilities (ID), so the prevalence of oral diseases does not change to any great extent for the ID population compared to the overall population with disabilities.

Zhou *et al* (2017) carried out a systematic review of the oral health status of young people with ID compared to populations without ID, which included data from 22 countries. They reported that children and adolescents with ID had fewer decayed and filled permanent teeth (DFT, $z=2.80$; $p=0.005$; $SMD= -0.36$, 95% CI -0.62 to -0.11). Zhou *et al* (2017) concluded that closer analyses of the data in the included studies in their review suggested that children and adolescents with ID, might have higher decayed, missing or filled surface (DMFS) scores, more missing permanent teeth due to dental caries and fewer

filled permanent teeth than their counterparts in a general population. The number of participants without an ID in this review was 16 times that of the number of participants with ID, 53,092 and 3,325, respectively, which may have reduced the ability to assess the true effect size, but may also be indicative of the difficulties of recruiting and examining children with ID.

However, children with ID are not a homogeneous group, they are not all equally at risk of developing dental diseases; considerable variation in dental caries experience has been found between groups with different levels of ID. For example, while 73% of 5-year-olds attending Irish schools for children with a moderate intellectual disability were caries free, 19% of them had a mean d3vcmft (visual dental caries) score of 4 or greater. Of these affected children, 8% had a d3vcmft score of 8 and 3% had a d3vcmft score of 9 or more. In the same study, 35% of the 5-year-olds examined had never been to a dentist (Whelton *et al* 2009).

In relation to the prevention and treatment of dental caries for children and adolescents with ID, the data show that, in general, teeth tend to be extracted rather than filled and fewer fissure sealants are placed (Hennequin *et al* 2008, Whelton *et al* 2009, Zhou *et al* 2017).

On the topic of gingival health, Zhou *et al* (2017) also reported that children with ID had significantly higher levels of dental plaque than those without ID ($z=2.62$; $p=0.009$; $SMD=0.67$, 95% CI 0.17 to 1.18) and the children with ID had significantly worse gingival status than those without ID ($z=2.53$; $p=0.011$; $SMD=0.63$, 95% CI 0.14 to 1.12). For this reason, more support generally with oral hygiene, more specific help with toothbrushing and more extensive periodontal treatment are required (Hennequin *et al* 2008, Whelton *et al* 2009, Zhou *et al* 2017).

1.4.3 Oral health of adults with intellectual disabilities

In 2003 in Ireland, adults with ID were shown to have greater levels of edentulousness and lower levels of natural teeth compared to the general population; as with the younger population with ID, treatment of dental caries by extraction is the more usual treatment provided (Crowley 2005).

This difference in the sequelae of dental caries in Ireland can be seen when comparing data from the 2003 survey of the population with ID and the 2002 survey of the general population in the table below (Crowley *et al* 2005, Whelton *et al* 2007) (Table 3). The prevalence of filled teeth for the ID population decreases with age while the prevalence of missing teeth increases; in the general population the numbers of filled teeth remains fairly stable.

Table 3: Sequelae of dental caries in Ireland

Population	Age Groups	Decayed	Missing	Filled	Total DMFT
Pop with ID 2003	16-34-year-olds	1.4	2.4	2.9	6.7
	55+ year-olds	1.0	26.7	0.4	28.1
General Pop 2002	16-24-year-olds	0.7	1.5	2.7	4.9
	65+ year-olds	0.5	22.8	2.6	25.9

Similar levels of caries and treatment provision for the populations with ID are seen in other countries (Anders & Davis 2010, Petrovic *et al* 2016).

In Ireland, between 16% and 52% of a population with ID were shown to have moderate or severe periodontal disease, the extent of the disease increased with age. Between 71% and 89% had calculus present (Crowley *et al* 2003). The methods used for assessing periodontal health in this latter survey were similar to those deployed in the Special Olympics assessment method; no probing of gingival tissues was undertaken. Comparing these data to the results from the 2002 survey of Irish adults, which used the more accurate CPITN assessment tool described earlier, between 0.4% and 12% had severe periodontal disease (deep pocketing ≥ 6 mm) and between 30% and 50% had calculus present (Whelton *et al* 2007).

As with the younger ID populations, higher and more severe levels of gingivitis and periodontitis are consistently seen in other studies or systematic reviews

of adult ID populations globally, using similar or more accurate assessment methods to those used in the Irish studies (Table 4) (Turner *et al* 2008, Anders & Davis 2010, Fernandez *et al* 2012, Petrovic *et al* 2016, Marks *et al* 2017).

Table 4: Gingival disease in ID and general adult populations

Study	Country	Data collected	Gingival Disease	Description used
Grey bars indicate general population groups (without ID)				
Crowley <i>et al</i> 2005	Ireland	2003	16% - 52%	Moderate to severe PD
Whelton <i>et al</i> 2007	Ireland	2002	0.4% - 12%	Severe PD (>6mm)
Dye <i>et al</i> 2007	USA	2004	10%	Moderate to severe PD (>4mm)
ARCPOH 2009	Australia	2004-2006	19.8%	% with 4+mm pocket depth
Turner <i>et al</i> 2008	UK	2005	63%	Gum inflammation
Fernandez <i>et al</i> 2012	USA	2005-2008	32%	Gingival signs
Marks <i>et al</i> 2018	Worldwide	2007-2015	34%-67%	Gingival signs

In summary, there is more untreated disease, more missing teeth (more often due to periodontal disease but also due to extractions being the more common treatment option provided to these population groups), less restorations, less replaced teeth and higher levels of moderate and severe periodontitis found in adults with disabilities than in the general adult population (Gabre *et al* 2001, Crowley *et al* 2005, Whelton *et al* 2007, Hennequin *et al* 2008, Australian Research Centre for Population Oral Health (ARCPOH) 2009, Anders & Davis 2010, Public Health England 2017).

Data from the Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing (IDS-TILDA) demonstrates the long-term outcomes of this deficit in the extent and type of care provided for people with ID. Older people (aged 50 or above) with ID in Ireland were shown to be more likely to be edentulous (34%) than those without ID (15%). Of the former, 61% were not wearing dentures (Mac Giolla Phadraig *et al* 2015). These trends are mirrored internationally (Mac Giolla Phadraig *et al* 2018).

In relation to manageability for dental care, more children and adults with disabilities may require general anaesthesia (GA) or sedation for general treatments or for complex or extensive treatments (Crowley *et al* 2005, Lader *et al* 2005, Hennequin *et al* 2008, Marcenes *et al* 2008, Anders & Davis, 2010). Effective prevention has to be the most appropriate way of reducing the need for this level of care (Crowley *et al* 2005).

1.5 Oral health interventions

A broad definition of oral health currently in use describes it as:

“multi-faceted and includes the ability to speak, smile, smell, taste, touch, chew, swallow and convey a range of emotions through facial expressions with confidence and without pain, discomfort and disease of the craniofacial complex.” (Glick *et al* 2016).

This definition expands to outline the three core elements of oral health: disease and condition status (which refers to a threshold of severity or a level of progression of disease), physiological function (such as the ability speak, smile or chew), and psycho-social function (the relationship between oral health and mental state) and acknowledges that oral health is influenced by determinants such as social and physical environments, health behaviours, and access to care.

Interventions designed to target oral health may aim to have an impact on some or all of the many facets of oral health.

1.5.1 Oral Health Interventions and Disability

Contributing to the complexity of describing an individual’s disability and its health impacts is the issue of determining their ability to care for their own oral health, within the context of their impairment, limitations and participation and within their social and personal environment.

Oral health interventions aimed at the general population might benefit people with disabilities but may not achieve the same outcomes. For example, the

physical beneficial effect of fluoride on enamel is likely to be the same for everyone, however, the level of exposure to the fluoride may differ between population groups. Similarly, interventions that consider the individual circumstances of populations, are more likely to be effective. For example, toothbrushing programmes may reduce plaque levels, but an intervention like this would be more likely to succeed in a population with disabilities if features such as reliance on others, physical aides and toothbrush modifications, are also considered as part of the intervention.

1.5.2 Evaluation of oral health interventions

To fully evaluate an intervention, it is important to take into consideration the process and impact of an intervention as well as the long-term outcomes. This requires detailed planning. It is important to know exactly what was intended and how it was expected to be achieved (Nutbeam 1998, Round *et al* 2005). A well outlined plan of action, as well as details of any tailoring to the particular context, should be reported, allowing the intervention to be properly delivered and ultimately replicated if desired (Craig *et al* 2008, Michie *et al* 2013).

In relation to what outcomes are achieved in an intervention, Kay & Locker (1998), in their review of the effectiveness of oral health interventions, were critical of the outcome measures used. Multiple methods exist to measure the same outcome, many do not meet quality assessment standards (Watt *et al* 2006). This continues to be an issue in current oral health research.

The importance of evaluating how, as well as what, was achieved is an important part of the evaluation process. Oral health research has been guilty in the past of focusing more on quantitative research. Qualitative research provides depth and insight into people's experiences, their social contexts; these elements may strengthen, support or diminish their health. The information gained from qualitative research may help explain the success or failure of an intervention (Nutbeam 1998).

This is not a new concept: O'Mullane (1976, O'Mullane *et al* 2012) reflected on complex oral health interventions and their settings and considered the

differences between explanatory trials (those carried out under ideal conditions) and pragmatic trials (those carried out in community settings), and the difficulties of evaluating them.

In the last decade, a large range of tools have been developed to assist designers in relation to ensuring their interventions are implemented, evaluated and reported in sufficient detail for transparent appraisal and replication if desired. Examples include the Precede – Proceed Model, AGREE II, the MRC framework, and PRECIS (Craig *et al.*, 2008; Brouwers *et al* 2011, Loudon *et al* 2013, Thorpe *et al* 2009). These tools should assist the design and evaluation of future oral health interventions.

Quality time must be spent on identifying the problem to properly design the solution and should take into consideration both the individual and the environmental factors that influence health. The active engagement of the intended participants has also been identified as a key element (Binkley & Johnson 2013).

1.6 Health behavioural change

The focus of healthcare has moved from the treatment of disease to the factors that support health and well-being, the effects of health-related behaviours, both positive and negative have been increasingly researched. There is now a greater understanding of how the determinants of health influence our health, determinants such as: genetics, where we live, our level of education and our support systems (Office of Disease Prevention and Health Promotion 2018). The salutogenic aspects of health are now considered to be the greater challenge: how can health behaviour be influenced?

Human behaviour is predominantly dependent on environment and largely controlled by antecedents and consequences. Health behaviour change is assumed to involve a variety of social, emotional and cognitive factors that work together (Schwarzer 2008). However, it has long been accepted that humans do have the ability to change their behaviour (Skinner 1969).

1.6.1 Behaviour change theories

For non-psychologists, it can be difficult to keep abreast of the most current or dismissed theories on behaviour change. A brief look back at the theories shows how they have developed over time, are linked to one another and how they have influenced current thinking (Appendix 1).

Given the multitude of behaviour change models in the literature, confusion may arise regarding which one to apply when attempting to understand or change health behaviour. Michie and colleagues have in recent times propelled thinking on behaviour change in an attempt to address this issue. In a seminal paper this team proposed that all interventions that are attempting to change behaviour must have a theoretical basis underpinned by evidence from the existing literature and to include a modelling phase, where the determinants of behaviour (the what) and the techniques to change these determinants (the how) are identified, before any intervention is implemented (Michie *et al* 2008).

Over a series of studies, Michie, Abraham and others have developed a list of the behavioural determinants and taxonomy of behaviour change techniques (BCTs), linked to the theoretical frameworks with which to influence these determinants. This taxonomy should assist non-psychologists and those new to the field of behavioural change, to identify the techniques in the literature even when the studies themselves fail to specifically identify them (Michie & Abraham 2004, Abraham & Michie 2008, Michie *et al* 2008).

In brief, the multiple theories of behavioural change overlap in many elements and some have built upon those that have gone before them. These theories have been used to explain behaviour rather than change it. By identifying the BCTs which elicit change in interventions, we may be better able to replicate them or include these elements when designing future interventions. The use of the BCT framework has been recommended by other behavioural experts in the oral health field (Asimakopoulou & Newton 2015).

Use of the BCTs may also broaden the criteria used to measure success, if an intervention can be shown to improve an individual's self-efficacy or have

increased their ability to plan around the barriers or failures, this can and should be regarded as success.

1.6.2 Behaviour change and people with ID

Our understanding of how a person with ID perceives health or how their health behaviour develops is not clear. People with ID experience more barriers than the general population in relation to leading a healthy lifestyle and making lifestyle changes, for example: intrinsic barriers relating to cognitive, behavioural and mobility impairments and external barriers such as financial issues, physical limitations and policy guidelines that limit health choices. These additional barriers require different approaches to be used or amendments to be made to existing general population interventions (Willems *et al* 2017).

Some small, qualitative studies have shown that people with ID have some understanding of what health entails and what constitutes a healthy lifestyle (Caton *et al* 2012, Kuijken *et al* 2016). Interviews with adults with ID in one study showed that they were largely unaware of dental problems and used appearance and absence of pain to judge the condition of their teeth (Cumella *et al* 2000).

The importance of support from carers, carers as role models, their dependence on caregivers for oral health decision-making and other environmental resources was identified in all these studies. The ability of the person with ID to communicate is essential when carrying out self-reported qualitative research on this population; this may mean that findings are only relevant to those with mild or moderate ID.

The use of conventional health behaviour change models when planning health interventions for this population is questionable, due to issues such as cognitive impairment as outlined earlier. People with ID may not have high levels of abstract thinking or be able to visualise hypothetical situations. They may not understand the long-term effects of risky health behaviour and so lack the motivation to change. They may not be able to develop coping strategies to the barriers encountered when attempting to change. The level of reliance on

informal and formal carers can affect freedom of choice and ability to act independently for people with ID. The influence of these carers on the health behaviour of people with ID must be taken into consideration (Brehemer-Rinderer *et al* 2014).

However, behaviour change techniques (BCTs) have been used in life style change interventions for people with ID with some success. A systematic review by Willems and colleagues (2017) on the use of BCTs on some life style change interventions (nutrition and physical activity) for people with ID, concluded that for best effect, researchers should consider testing particular BCTs to see how they might be made to work for people with ID, for example with support from carers, and careful consideration should be given during the design phase as to which and how many BCTs should be included.

1.7 Rationale for this research

The oral health of people with ID is poorer than the general population. There have been many research studies assessing the oral health status of people with disabilities; these studies have been systematically reviewed resulting in a better picture of what elements need to be addressed, in order to reduce the inequalities in oral health status between people with disabilities and those without (Moreira *et al* 2016). People with ID evidently require more support in maintaining their oral hygiene.

However, the research to date, in relation to the actual oral health interventions, and their outcomes, which have been undertaken for a population with disabilities, is not clear. There is little transparency as to what types of disabilities have been targeted, what types of interventions have been undertaken, what outcomes have been assessed and what are the contexts and mechanisms that facilitate or create barriers in relation to the success of these interventions.

Oral health interventions aimed at improving the oral health of people with disabilities may need to use different approaches depending on who the intervention is targeting. Carers of people with physical disabilities and people with mild ID may respond to interventions that improve oral hygiene skills or

incorporate BCTs, while introduction and monitoring of oral healthcare policies and processes within institutions may be of greater importance for people with more severe levels of ID. In order to be properly evaluated and to allow replication, interventions that incorporate behavioural change must have a theoretical basis, be evidence based and must identify the determinants of behaviour (the what) and the techniques to change these determinants (the how) involved in the intervention. The type and success of the existing oral health related interventions for people with ID is not known. In order to ensure that future interventions will improve the oral health of people with ID, in a sustainable way, a review of the existing interventions is required as a starting point.

Chapter 2 – General Methodology

2.1 Introduction

Decisions regarding healthcare interventions for individuals or for healthcare policy should be informed by the best available evidence. The volume of research available may make finding the best evidence difficult, the quality of the evidence can vary, studies may have conflicting findings (Abalos *et al* 2001). A source that has considered and analysed all the evidence in a systematic way can improve the decision-making process for healthcare professionals seeking the best evidence to inform their practice and thus the health outcomes of the people they care for (Abalos *et al* 2001, Centre for Reviews and Dissemination (CRD) 2008).

The systematic review has long been considered the most effective way to synthesise the findings of multiple studies to make evidence-informed recommendations on the effectiveness of a specific intervention or treatment option. A Cochrane Review is regarded as the gold standard for systematic reviews (Higgins & Green 2011). A protocol clearly sets out the parameters that will be used to assess the intervention of interest before the systematic review is undertaken, this minimises subjectivity and potential bias. Both the content and infrastructure of the data from the studies are minutely examined in relation to the definitions, study design and methodologies used, and quality assessed. All of this is systematically recorded so that the process is reproducible (Bartolucci & Hillegass 2010).

Despite the high esteem in which systematic reviews are held, they do have their limitations. Randomised controlled trials (RCTs), the building blocks of a systematic review, are criticised for not being applicable in the “real world”. Studies of complex interventions need to detail the processes and mechanisms by which the outcomes were achieved to provide useful information (Pawson 2006, Kastner *et al* 2012).

There has been an increase in the different types of evidence reviews being undertaken, that seek to capture the rich contextual details that may be lacking in the conventional systematic reviews (Kastner *et al* 2012, Tricco *et al* 2016a,

Tricco *et al* 2016b). These reviews focus on developing theories that allow a greater understanding and insight into the effectiveness of interventions. This knowledge is being used to inform practice and policy (Pawson 2013, Pawson *et al* 2014).

Questions about how and why an intervention works are increasingly regarded as being just as important as what works. However, in the dental field, we have been slow to embrace this thinking (Stewart *et al* 2008, Petticrew *et al* 2015). In the past, dental professionals tended to report qualitative data in an informal way, often it was simply the observations of the research team and unlikely to have been part of the formal research process. In the last decade, the dental profession has begun to realise the rich source of data qualitative research can unearth, increasing our understanding of how our patient's life experiences and environment may impact their oral health behaviour; how they understand or view their oral health (Stewart *et al* 2008).

When searching for both quantitative and qualitative data, different search methods are preferable. The databases for qualitative studies are not as developed as the quantitative databases. The titles and abstracts of qualitative articles use more descriptive terms and are not as structured as their quantitative counterparts. A search for quantitative data may identify some associated qualitative studies in the course of the search, but the results tend to be random rather than systematic. A similar situation would arise if the reverse search was undertaken. If a combined qualitative and quantitative search is undertaken, the search terms must be much broader, resulting in large numbers of studies to be screened, decisions regarding inclusion or exclusion may require much more full text reading due to the unstructured format of the titles and abstracts (CRD 2008).

Policy makers are increasingly demanding that reviews of current evidence to inform best practice are completed in shorter time frames; conducting rigorous and transparent systematic reviews are very time consuming. Many of these newer review methods acknowledge that the price of speed is at the cost of comprehensiveness (Grant & Booth 2009). However, this may not be a bad thing, for example, a Realist review does not set out to find all the evidence on

a topic, but to find enough evidence to support or reject the identified theories. The search is purposive, using specific search terms relevant to the theories. Once saturation has been reached, when it is reasonable to claim that the theory is coherent and plausible, then the search can stop (Rycroft-Malone *et al* 2012, Wong *et al* 2013).

Additionally, it is important that the findings of reviews can be implemented. There is increasing evidence of a knowledge-to-action gap, the translation or implementation of the knowledge gained from reviews needs to be presented in an accessible format so that healthcare professionals can see how it can be implemented in practice (Graham *et al* 2006, Wallace *et al* 2014). Cochrane reviews include a “plain language” summary and a concluding section on the implications for practice and research. Qualitative reviews such as Realist Reviews present their findings in a format that can be practically applied in the real world. The intention of a Realist review is to explain how and why an intervention worked in a particular context. This makes it easier for a healthcare professional or policy maker to see if the intervention is likely to work in their context, or what adaptations may be needed for it to work in their setting (Wong *et al* 2013, Fletcher *et al* 2016).

2.2 Choosing multiple review methodologies

The choice of review types used in this research came about gradually and was made in reverse order to the order in which they are presented in this thesis. An awareness of the complexity of oral health interventions for people with disabilities was the starting point. A qualitative review was felt most appropriate to unearth the contexts and mechanisms that influenced the outcomes of these complex interventions. However, the focus of a qualitative review, given the nature of the data, needs to be fairly narrow. After some discussion and reflection, it soon became apparent that for the qualitative review to be useful, it would be best if it focused on an intervention that was likely to have positive clinical outcomes, which tend to be measured quantitatively.

The need for a systematic review of the quantitative outcomes then became obvious, as no systematic review of oral health interventions for people with disabilities had been completed. In discussions with the Cochrane Oral Health Group in relation to the title or focus of a systematic review of this population, they suggested a Scoping Review of the types of interventions that have been carried out on the population with disabilities to help identify the focus. It was the findings of the Scoping Review that determined the focus of the Cochrane Review, which then determined the focus of the Realist Review. Initially, it was thought that focusing on interventions targeting children and adolescents with disabilities for the Scoping Review would be productive, as the effects of an intervention targeting this age group would impact on the participants into the future and additionally, the search would be manageable. However, when the Scoping Review was completed, it was clear that it would be necessary to include all age groups in the Cochrane Review.

When defining the type of oral health interventions relevant to the reviews in this research, I commenced with a broad definition for the scoping review, narrowed the definition for the Cochrane Review and further narrowed it for the Realist Review. The three review methodologies will be described in this chapter. The specific methods used in each of the reviews will be detailed in their own separate chapters.

2.3 A Scoping Review

The most common form of scoping reviews, is a “literature map”; it constitutes a practical, methodological approach to finding relevant literature in a previously un-searched subject area, it spells out the origins of work on the subject and gives a good feel for its chronological development (Arksey & O’Malley 2005, Anderson *et al* 2008).

Given the limited information available on the types of oral health interventions implemented for those with disabilities, we felt that this method was well suited to finding any relevant literature on the topic.

Traits that distinguish a scoping review from a systematic review include the lack of a defined research question, the knowledge that this is a broad

approach, as well as the inclusion of all study designs, without intent to assess the quality of the studies, or to draw specific conclusions. The purpose is to quantify the elements of the studies, such as the populations, type of interventions and outcomes; and ultimately, to use this information to develop and refine a systematic review question that will be feasible as well as having sufficient literature to review (Arksey & O'Malley, 2005, Levac *et al* 2010).

The steps in a Scoping Review as outlined by Arksey and O'Malley (2005) (Table 5), are similar to a systematic review and should be as rigorous and transparent.

Table 5: Steps in a Scoping Review (Arksey & O'Malley 2005)

	Steps in a Scoping Review
1	Identifying the research question
2	Identifying relevant studies (Inclusion and Exclusion Criteria)
3	Study selection
4	Charting the Data (Data Extraction)
5	Collating, summarizing and reporting the results
6	Optional stage: Consultation Exercise

2.3.1 The focus of the Scoping Review

The research question for the scoping review was: What is known from the existing literature about the oral health interventions that have been implemented for children and adolescents with disabilities, specifically, the populations targeted, the types of interventions implemented, and the outcomes measured?

For the Scoping Review, the definition of an oral health intervention used was: any action that would impact on oral health, specifically, prevention of dental caries and periodontal disease. This would include interventions in relation to risk assessment, screening, treatment, prevention, education and training; public health initiatives; environmental initiatives; hospital, institutional, clinic and home-based initiatives; charged or free initiatives. The populations included in the Scoping Review were children and adolescents with disabilities, their parents, carers, teachers and healthcare workers.

The findings of the scoping review have been published (Waldron *et al* 2016) and are presented in Chapter 3.

2.4 A Cochrane Review

A systematic review gathers all available evidence, meeting a pre-determined set of criteria, to answer a specific question. A systematic review reduces bias by following a systematic approach and ultimately results in findings that are more reliable than individual studies on their own, from which conclusions can be drawn and decisions made. The Cochrane Collaboration have, for the last 25 years, set themselves the mission of promoting evidence-informed health decision-making by producing high-quality, relevant, accessible systematic reviews and other synthesized research evidence. Their work is internationally recognized as the benchmark for high-quality information about the effectiveness of health care (Higgins & Green 2011).

Cochrane reviews, and protocols for reviews, are prepared in the Cochrane Collaboration's Review Manager (RevMan) software, have a uniform format and are prepared by teams. There are 53 Cochrane Review Groups, each covering a different topic, one of which is The Cochrane Oral Health Group (COHG). The title of a Cochrane Review is agreed after discussion with the relevant group and submission of a title registration form. This form contains details such as the motivation for the review, the types of studies to be reviewed and the team of authors involved. Once the title has been accepted, the review team are invited to submit a protocol for the review. The protocol and the review are

scrutinised by the editorial team in the Cochrane group as well as subject to external peer review before either is published on the Cochrane website.

The sections of a review are the main text (Background, Objectives, Methods, Results, Discussion and Conclusions), the tables (Characteristics of the included studies tables, Risk of bias tables, Excluded, awaiting classification and ongoing studies tables, Summary of findings tables and Additional tables), Studies and References, Data and Analyses (Any forest plots from meta-analyses) and Figures.

2.4.1 The focus of the Cochrane Review:

The focus of the Cochrane Review was determined by the findings of the Scoping Review (Waldron *et al* 2016), which identified that there were sufficient studies on populations with ID targeting oral hygiene interventions to warrant a systematic review. This process of determining how the review question was chosen is described in more detail in Chapter 4.

For the Cochrane Review, the interventions of interest were oral hygiene interventions, which were defined as: any intervention that may impact on the duration, frequency and/or effectiveness of oral hygiene practices. The populations included in the Cochrane Review focused on two distinct groups: people with an ID and the carers supporting, assisting or providing oral hygiene care for them.

The objective of the Cochrane Review therefore was: To assess the effects (benefits and harms) of oral hygiene interventions for people with intellectual disabilities (ID). The primary outcome was the gingival health of people with ID, the secondary outcomes were: Oral hygiene knowledge acquisition by people with an ID or the acquisition of knowledge by their carers with regard to oral hygiene; Changes in behaviour, attitude and self-efficacy of people with an ID or their carers; Quality of life changes or long-term, patient-centered outcomes for people with an ID (e.g. tooth retention); Dental caries levels of people with an ID and any unintended (positive or negative) effects of the interventions.

2.5 A Realist Review

As an introduction to the concept of Realism, the term “hypothetical” comes to mind when trying to describe the methodology. It requires the reader to take a leap of faith, in order to discover that which cannot be seen in order to understand how and why things work. Realists do not produce conclusions following their research, but develop theories or hypothesised causal pathways, in relation to what they think may be the explanation for particular mechanisms or contexts influencing particular outcomes.

Realism, broadly speaking, sits between positivism (‘there is a real world which we can see and understand directly through observation’) and constructivism (‘given that all we can know has been interpreted through human senses and the human brain, we cannot know for sure what the nature of reality is’) (Pawson 2006). Realists seek to look deeper beyond the limits of empiricism, which maintains that in order to determine something it must be observable; be able to be experimented upon or experienced; with attention to variables. Realists seeks to gain a more productive understanding of what they see in the real world, to understand the underlying power that links the variables and explains what causes them to be connected. They aspire to develop theories about this through their perceptive imaginative thinking (abduction/retroduction).

Realists do not make conclusive statements, because they maintain that all the mechanisms or contexts cannot be fully identified, that there are infinite variations of mechanisms and contexts, they can be invisible, immeasurable, or obscure (Pawson 2006). A realist review, potentially, would facilitate identification and examination of the theory, contexts and mechanisms within interventions which might positively influence the outcomes. In order to identify causal connections, it is necessary to understand outcome patterns, or as Pawson (2006) refers to them “Demi- regularities”, rather than seek outcome regularities.

However, lest this description sound too abstract, the methodology is guided by methodological guidance, publication standards and training materials for realist and meta-narrative reviews: Realist And Meta-narrative Evidence

Syntheses: Evolving Standards [sic](RAMESES) and training materials, which have been followed in this review (Wong *et al* 2013).

The use of realist methodology alongside a more traditional systematic review to study the effect of oral health interventions will allow a deeper synthesis of how and why an intervention does or does not work as well as what effect it has. RCT's have an ability to determine what effect the intervention has when a number of static variables are applied but may not always be able to identify how and why it worked (Fletcher *et al* 2016).

Given the complex nature of oral health interventions in relation to the variety of mechanisms and contexts, which are complex processes as opposed to static variables, many of which will be unobservable with multiple possible outcomes likely, it is important that these underlying causal pathways, are also considered to ensure that the intervention can be repeated with consistent outcomes when the mechanism and/or contexts may vary to any degree. Any theory developed from these observed and unobserved elements will need to be tested and retested in future studies and are likely to evolve and undergo revision over time, they are after all, theories.

The focus of the Realist Review was determined based on the early findings of the Cochrane Review and with guidance from Realist Review experts during training workshops on Realist methodology. The extensive involvement of carers in the oral hygiene care of people with ID, particularly those with severe or profound levels of ID, and the impact of the level of care they provided, on the oral health of the people with ID for whom they care, was evidenced in the Cochrane Review, and identified by the review team as an important area for further review. The likely volume of data gathered in a Realist Review and the time available to complete this review, also influenced the focus.

The interventions of interest were carer-led oral hygiene interventions, which were defined as: any carer-led/ carer-implemented intervention that may impact on the duration, frequency and/or effectiveness of oral hygiene practices for people with intellectual disabilities or their carers.

2.5.1 Why a realist approach was undertaken

A basic assumption of realist evaluation is that programmes are complex interventions introduced into complex social systems (Pawson 2013).

The outcomes from oral health interventions involving behavioural change have been very unpredictable. Interventions have worked in one setting and not in another or shown benefit in the short term but not long term (Kay & Locker 1998, Owens 2011). The need to develop theories about how and why an intervention might work is increasingly recognised. This is particularly important in situations where the intervention is attempting to assist people to change their behaviour and where the individual's motivations and setting may influence the outcome. Thus, the limitations of the more traditional systematic reviews, when investigating the success or failure of complex interventions such as these, are increasingly evident (Kastner *et al* 2012).

In eliciting change, there is an underlying process that takes place, involving interaction, persuasion and reasoning. It is essential to gain an understanding of the contexts: the resources, culture and opportunity structures that might influence the outcomes. The vast array of contexts that might arise in any given intervention cannot practically be reduced or eliminated, some contexts will enable, some will limit, but they all must be considered in the intervention theory. This is particularly true in the field of oral health promotion, where there is no clear best practice and the complexity of intervention designs, methodology and outcomes are vast. For a population with ID, the implementation and fidelity of interventions may, additionally, be compromised by any number of issues such as the range of disability, the cooperation of the participants, or the resources to hand. Evidence regarding the elements of an ideal intervention would be valuable.

Thinking through how each issue applies to the interventions about to be investigated will help in the identification of the theories about when, why and how the interventions work. Thus, the key characteristics of complexity in interventions can be considered under the Realist complexity acronym – VICTORE (Volitions, Implementation, Contexts, Time, Outcomes, Rivalry and

Emergence) as outlined by Pawson in "The science of Evaluation: a realist manifesto" (Pawson 2013).

The acronym stands for:

Volitions: the "choice architecture" of a program including how program subjects might respond to a program or intervention;

Implementation: the implementation chains of an intervention/program which are prone to inconsistency and interpretation, blockages, delays, and unintended consequences;

Contexts: the context of an intervention refers to the circumstance in which it plays out. Pawson outlines a "four I's" framework: Individuals (characteristic and capacities of stakeholders in the program); Interpersonal relations; Institutional settings; and Infrastructure (which refers to the wider social, economic, and cultural setting of a program/intervention);

Time: the history and of timing of an intervention;

Outcomes: approaches for monitoring and ways stakeholders might interpret the outcomes;

Rivalry: the pre-existing policy landscape in which the program is embedded – this primarily refers to other, contiguous programs and policies may share or oppose the ambitions of the intervention under study and actions of stakeholders and subjects under study; and

Emergence: potential emergent effects, long-term adaptations, and unintended consequences associated with the program/intervention.

The key characteristics of carer-led oral hygiene interventions as considered under the acronym VICTORE were as follows:

- The role of carers in these interventions are influenced by the level of **Volition** they demonstrate when making choices in relation to their involvement in the oral hygiene care of the people with ID for whom they care. Their level of interest being one of the most important, as it will impact on all other theories. Changing mindsets takes time; the programme 'architecture' must give the carers the time to make the choices that result in developing an interest in the topic.
- The **implementation** chain of the intervention needs to be considered to fully understand how it impacts on its success or failure. It is

important to consider the stages at which modifications might be made to the original concept for the intervention and the reasons for them. Often changes can be related to financial aspects or resources, how do these changes impact on the outcomes? If there is insufficient staff to carry out the daily oral hygiene practices, then the intervention is likely doomed to failure.

- Interventions are influenced by an endless source of **contexts** which can, for convenience, be grouped under the four I's as outlined by Pawson (2013)
 1. Individuals – the characteristics and capacities of the various stakeholders in the programme
 2. Interpersonal relations – the stakeholder relationships that carry the programme.
 3. Institutional settings - the rules, norms and customs local to the programme
 4. Infrastructure – the wider social, economic and cultural setting of the programme.

For example, in this instance, the educational level of the carers, the level of support from management or other healthcare professionals and colleagues, any goals or incentives present and the financial resources available.

- **Time** spent on training, time available to carry out the oral hygiene practices, time of follow up support, monitoring or incentivisation, and length of time the intervention is evaluated may all impact on the success of the intervention.
- Measurement and analysis of both qualitative and quantitative **outcomes** may improve the findings of the realist review.
- Competing forces such as other commitments of staff or carers, other health priorities of the people with ID, may create **rivalry** to the success of the intervention.
- Unexpected or unintended outcomes, contexts or mechanism may **emerge** which will need to be considered. Some consideration at the outset will need to be given to the focus of the review and if these

unexpected outcomes can be assessed with the outcome measures identified for the review.

Tricco *et al* (2016b) identified nine knowledge synthesis methods that can be used to generate or refine theory in relation to how health interventions work: concept synthesis, critical interpretive synthesis, integrative review, meta-ethnography, meta-interpretation, meta-study, meta-synthesis, narrative synthesis, and realist review. Of these, a realist review was identified as having clear guidance on all steps of the process, being unique in its ability to combine quantitative and qualitative evidence and having the ability to identify or refine theories that can be applied in health care settings as well as the potential to influence policy (Tricco *et al* 2016b).

Pawson & Tilley summarised what they felt to be the crucial elements of realist research as follows “outcomes unearthed in empirical investigation are intelligible only if we understand the underlying mechanisms which give rise to them and the contexts which sustain them” (Pawson & Tilley 1996).

The use of a Realist review alongside a Cochrane review may unearth useful evidence in relation to what interventions work for whom and under what circumstances. This may allow for practical recommendations to be made in relation to how interventions should be implemented in different settings and ultimately influence policy (Pawson *et al* 2005, Greenhalgh *et al* 2007).

2.5.2 The focus of the Realist Review

The research question for the Realist Review was “What is it about carer-led oral hygiene interventions for people with intellectual disabilities that works and why?”

The aim of this Realist Review was to develop a conceptual model of what carer-led oral hygiene interventions for people with ID should look like, by identifying the contexts and mechanisms within these types of interventions, that influence the outcomes, implementation and sustainability of the interventions.

Chapters 4, 5 and 6 will outline in more detail, the interventions and outcomes assessed in each review.

Chapter 3 A Scoping Review

3.1 Introduction

The purpose of this scoping review was to identify the types of interventions that have been implemented for children and adolescents with disabilities, specifically in relation to the populations targeted, the types of interventions implemented, and the outcomes measured. It was decided to limit this review to children and adolescents to ensure that the search would be manageable; it was also felt that the effects of any intervention targeting this age group would have the potential to continue to impact on the participants into the future.

3.2 Methods

3.2.1 Inclusion and Exclusion Criteria

Our intention was to complete as comprehensive and inclusive a search as possible while being cognisant of making the search a manageable one. Therefore, the search terms used were broad and there were no time limits placed. However, the search was limited to an electronic search of five databases (Pubmed, Embase/Medline, PsycInfo, Scopus and Cochrane). In developing the search strategy, key terms were considered and discussed with those experienced in the oral health and special care fields, an experienced Cochrane systematic reviewer and subject librarians. Known relevant articles were consulted and key terms checked. The PICO (population, intervention, control, and outcomes) method for framing a researchable question was used.

Participants

For this review, the studies to be included must have children or adolescents (from birth to 18 years) as participants, with disabilities (CAWD), as currently defined by the International Statistical Classification of Diseases and Related Health Problems (ICD10) (WHO 2011). In addition, they must have required some assistance with their oral hygiene and general health care, and have received the intervention either at home, in day care, in hospital or in residential care settings.

Interventions

All oral health interventions that aimed to improve the oral health of the participants, or directly assist parents, carers or healthcare professionals (HCP) in supporting the participants to improve the oral health of CAWD, were considered under the following headings:

- Oral health related educational interventions including education about the development and anatomy of the teeth and gums, care of the teeth and gums, diseases of the teeth and gums and prevention of diseases of the teeth and gums.
- Oral health related behavioural and psychological interventions including motivation in relation to tooth brushing routines and dietary habits.
- Oral health related skills training for paid and unpaid carers, including interventions assessing the oral health status of the participants, tooth brushing skills, assisting the participants in relation to tooth brushing, designing and implementing oral health related routines or policies in different settings.
- Oral health related skills training for participants, including interventions in relation to tooth brushing skills and routines and dietary choices and routines.
- Oral health related clinical and pharmacological interventions, including intervals between dental examinations, application of topical fluorides, fissure sealants, oral prophylaxis, oral hygiene aids and type and strength of solutions.
- Oral health promotion interventions including systemic fluoridation, international, national and local community policies.

Outcomes

The interventions must have included a comparison to usual, no care, or comparable interventions. Intervention outcomes including quantitative indicators such as indices recording: Plaque score, DMFT/dmft, Bleeding on probing (BOP), Community periodontal index of treatment needs (CPITN) or qualitative measurements such as those assessing the knowledge, attitudes and skills of the participants, parents or carers, or using an assessment that can be compared to one of these indicators, were included in the review. The following databases were searched: Pubmed, Embase/Medline, PsycInfo, Scopus and Cochrane. No specific ethical approval was required for this study

as the secondary analysis of the primary research in this scoping review was limited to information already in the public domain. However, every precaution was taken to ensure the continued anonymity of the participants and that the results were used in the manner to which the subjects had originally consented.

3.3 Study Selection

The studies were examined by two reviewers, one of whom is an expert in the field of Special Care Dentistry (CMGP) and the other is a dental health care professional with experience in Health Promotion (CW). The studies were reviewed firstly by the titles and abstracts. Details such as keywords, authors, publication and institutions, were available to the reviewers. The protocol used for screening is outlined in Table 6.

Table 6: Screening protocol

The study must meet all of the following four criteria which must be identified within the title or abstract: ☐ The study reported must relate to Oral Health ☐ The study reported must be focused on a population with disabilities as defined by the ICD ☐ The study reported must include infants, children and/or adolescents (0-18 years) ☐ The study reported must involve an intervention to broadly impact on oral health Each reviewer should determine each article to be “relevant” (Y), “not relevant” (N) or “unsure” (?) independently and to be overly inclusive at this stage as per the Cochrane standards. Each of the four criteria was also to be recorded as present (Y) or absent (N) or unclear (?). Screening Protocol

The search terms used for PubMed, which was adjusted as required for the four other databases, PsycInfo, Embase/Medline, Scopus and Cochrane Library are provided in Table 7.

Table 7: Scoping Review search terms used in the PubMed database

DATABASE	SEARCH TERMS USED
PubMed	(child* OR infant OR adolesc* OR teen* OR "young adult") AND (disabil* OR "Vision Disorders" OR "Communication Disorders" OR "special needs" OR retard* OR impaired OR "Disabled Persons" OR blind OR deaf OR autism*) AND ("mouth diseases" OR "dental diseases" OR "tooth diseases" OR dental decay OR dental caries OR "gum disease" OR "gum problems" OR "dental health" OR "oral health") AND ("Health Services" OR "Public Health Dentistry" OR "Risk Reduction Behavior" OR "Health Behavior" OR "Health Policy" OR "Occupational Therapy" OR "Education, Special" OR "Health Education" OR "Early Medical Intervention" OR "Early Intervention (Education)" OR dentistry OR "preventive dentistry" OR "dental care" OR "dental care for disabled" OR "oral hygiene")

Pilot testing, discussion and revision of the process were undertaken. As a measure of reliability between both reviewers, a kappa score (0.81) of the inter-rater agreement was undertaken and found to be acceptable based on a screening of 430 of the identified studies. The bibliographic software Endnote was used to record and manage the articles.

3.4 Screening

A total of 3,909 articles were identified in the search of the five databases, which was completed in August 2015. A total of 1,257 duplicate articles were excluded using Endnote (575 articles) and hand screening (682 articles). A total of 220 articles were written in 17 non-English languages: Chinese, Croatian, Czech, Danish, Dutch, French, German, Greek, Hungarian, Italian, Japanese, Korean, Norwegian, Polish, Russian, Spanish, and Turkish. These articles were screened by title and abstract, 12 were considered to be relevant and in another 23 the relevance was unclear. However, we felt that this small number would not impact on the overall findings of this scoping exercise and so it was decided, due to the cost implications of acquiring translations, to exclude them.

Following screening of the remaining 2432 articles by title and abstract, 2,179 articles were excluded as they did not meet the inclusion criteria. Of those excluded, 1,551 had no intervention, 606 did not include participants with a

disability, 17 did not include children or adolescents and 5 were not on the topic of oral health, or any combinations of these criteria.

Eighty-one articles were considered to have interventions of interest and a further 170 articles required full text reading to determine if they should be considered; 87 of these had no abstract. The full articles were accessed from the Dublin Dental University Hospital Library, Trinity College Library or requested via the inter-library loan system. All, except one article (Baker, 1969 in the Illinois Dental Journal), were sourced. A further 139 articles were excluded at this stage for the following reasons: no intervention (105), no disability (20), adults only (6), case reports (7), article not found (1). This left 112 articles which met all the inclusion criteria. Figure 1 represents the modified PRISMA (Preferred Reporting Items for Systematic Reviews and Meta Analyses) flow diagram of this process. PRISMA is the evidence-based, minimum data set of items for reporting in systematic reviews and meta analyses (Moher *et al* 2009).

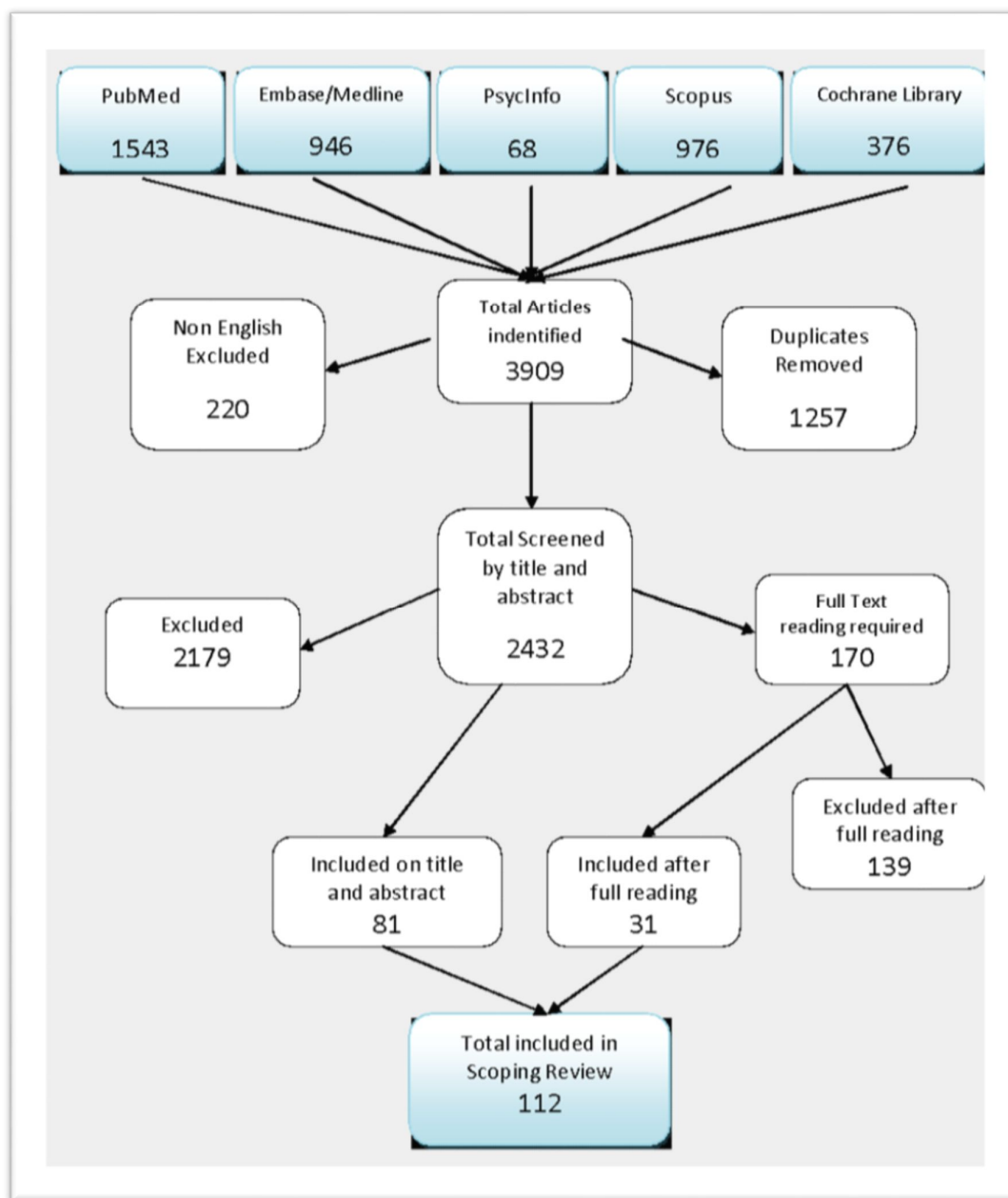


Figure 1: Prisma Flow Chart for the Scoping Review

3.5 Data Extraction

Reading of the abstract and/or the full text of the remaining 112 articles was undertaken, with the purpose of identifying the following:

- Which population of children or adolescents with disabilities were targeted
- If others such as parents, carers or HCP were involved
- What type of intervention was implemented

- What outcomes were measured

We recorded these details for each article in an excel spreadsheet. Any changes over the years, in the types or focus of interventions included in this scoping review, were also examined.

3.6 Results

3.6.1 Participants

The types of disabilities were clearly identified for the most part, although some were grouped by a broad disability such as physical disability, whilst others were very specific, such as Cerebral Palsy (Table 8).

Table 8: Types of disabilities identified in the interventions

Type of Disability	Number of Articles
Autism	6
Blind	9
Cerebral Palsy	3
Deaf	3
Down Syndrome	12
Intellectual Disability	32
Mixed Disabilities	26
Physical Disability	8
Other	4
Unclear	9
Total	112

A wide variety of persons other than children or adolescents with disabilities (CAWD) were involved in the interventions, either as educators, supervisors or deliverers of care, or as the target of the interventions (Table 9).

Table 9: Groups other than CAWD involved in the interventions

Groups other than Children and Adolescents with disabilities involved in the intervention	Number of articles
Carers	20
Dental professionals or students	9
Medical staff, Nurses, Occupational Therapist	5
Parents	11
School/ Teachers	2
Total	47

3.6.2 Interventions

For analysis, we grouped the types of interventions under the broad headings of Education, Skills, Therapy or Complex.

Interventions that purely involved education (N=8) were mostly targeted at the populations other than CAWD, i.e. carers and parents. The outcomes measured for these interventions focused mostly on knowledge, understanding and attitudes.

Skills based interventions were predominately focused on tooth brushing skills (N=16), for both CAWD and their carers but also included the development of skills in behavioural techniques and skills in assessment of oral health for the carers.

Therapy-based interventions included those aimed at improving the opportunities to provide care or provision of care (N=8), provision of particular types of therapy such as Chlorhexidine (N=7) mouth appliances (N=5), fissure sealants (N=3), fluorides (N=3), Botulinum toxin (N=3) or comparisons of different aids and solutions (N=13) or some combinations of these therapies. Complex interventions incorporated more than one element. The majority of the interventions were therapy-based interventions (N=52), with pure educational interventions being the least common (Table 10).

Table 10: Types of interventions

Intervention Type	Number of articles
Complex	21
Education	8
Skills	31
Therapy	52
Total	112

3.6.3 Outcomes

The outcomes addressed are shown in Table 11. A number of indices were used to measure the same clinical condition. Detailed examination of the indices would need to be undertaken to determine if the results could be compared in a meta-analysis.

Table 11: Outcomes measured

Outcome measured	Number
Plaque	47
Gingivitis	36
Dental caries	19
Attitude, knowledge, Self-efficacy	10
Bacterial counts	10
Orthodontic results/ oral function	5
Periodontal pocket depths	5
Saliva	4
Compliance	4
Quality of life	3
Anxiety	2
Others	14
No outcome measure identified	13

3.6.4 Timeline

The earliest published intervention found was in 1965. All interventions types were spread evenly between the 1960's and 2010's except for a lull in skills-based interventions in the 1980's and 1990's, with only two of the 31 skills-based interventions occurring in these decades, the remainder occurred equally in the decades before (14) and after (15). There has been a noticeable increase in the number of interventions reported in the last 15 years, 2001-2016 (Figure 2).

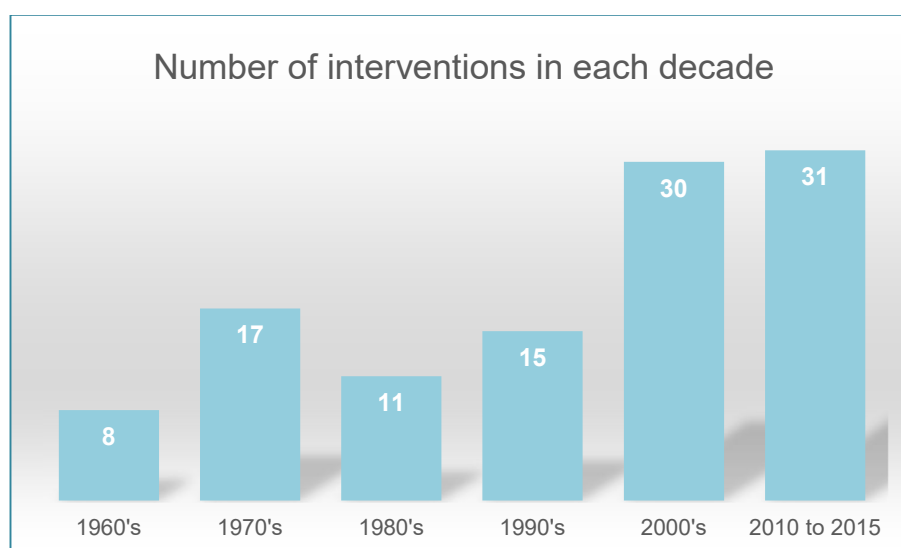


Figure 2: Number of interventions over the decades

3.7 Discussion

A relatively small number of published oral health interventions (N=112) for children and adolescents with disabilities met the criteria outlined in this scoping review; the earliest identified was reported in 1965. The number of interventions implemented and evaluated has increased dramatically in the last 15 years (2001-2016); 54% of the interventions reviewed were reported in this time period.

The most commonly targeted group in these interventions was people with an intellectual disability. Usually it was carers and parents who were commonly involved with the interventions, both as deliverers or supervisors of the care

or as the target of the interventions. Carer or parent-led interventions are common in general health interventions also, both for those with and without disabilities (Niemeier *et al* 2012, Hithersay *et al* 2014). The benefit of using carers or parents in health interventions is unclear, they appear to affect the outcome of interventions both positively and negatively; focusing on skills rather than knowledge acquisition, for carers and parents, may be more useful and it can be a cost-effective way of delivering the care that is required (Hithersay *et al.* 2014).

The most frequently cited type of intervention was therapeutic; the range of therapies was diverse. The next most frequently published intervention type was skills-based interventions; these predominantly focused on tooth brushing skills for both children and adolescents with disabilities and their carers.

The Scoping Review was helpful in determining the topic for a systematic review, it identified the limitations of basing a systematic review on the intervention setting or carers involved, since this was likely to result in few, comparable studies being identified. Other limitations were likely if specific age groups of those with disabilities were targeted, for example, children or adolescents; the range of Intelligence Quotients (IQ) for the various age groups depending on their disability was likely to be complex.

Splitting the entire population by disability type was a possibility, as the studies seemed to focus on a selection of identified disabilities. However, currently, researchers are encouraged to use the ICF definition of disability (WHO 2001), which focuses on the health conditions, the environmental and the personal factors of the individual rather than labelling the disability *per se*. The definition of the disability to be reviewed should consider this element carefully. However, it was also considered possible to split the entire population by intervention types, such as specific therapeutic or skills-based interventions.

3.7.1 Limitations

The decision to limit the participants in the Scoping Review to children and adolescents, was made for reasons of manageability as well as a belief that

focusing on interventions targeting children and adolescent would be productive, as the effects of an intervention targeting this age group would impact on the participants into the future. However, based on the amount of data identified in the Scoping Review, it was felt that all age groups should be included in the planned Cochrane Review. The findings of the narrower Scoping Review worked as a basis for this Cochrane Review, as most of the interventions identified in the Cochrane Review involved adult participants. However, in hindsight, including all age groups in the Scoping Review, would have been more thorough. A future scoping review of oral health interventions, targeting adults with disabilities, may reveal different findings on the disability populations targeted, the types of interventions implemented, and the outcomes measured.

The limitations of scoping reviews in general, and this review specifically, is that they are normally based on a reading of the title and abstract only for the majority of the studies, this does not reveal sufficient details of the study designs used. A more detailed analysis of this element may be beneficial, but more time consuming (Arksey & O'Malley 2005).

3.8 Conclusions

The use of a scoping review of children and adolescents with disabilities, as well as achieving its objectives of identifying the range and types of oral health interventions, the disability populations targeted and the outcomes assessed, was also found to be an efficient and effective way of supporting the development of a research question for a more focused systematic review on this topic.

The population most commonly targeted in the interventions was those with intellectual disabilities, who are recognised as a group with poorer oral health than general population groups and who require more support in maintaining their oral hygiene. This makes a strong argument for focusing on this population for a Cochrane Review, in order to assess the effectiveness of the interventions targeting them. Given the relatively small number of interventions identified in the scoping review, with no assessment of the quality

or study design, it made sense to expand the population from children and adolescents to all age groups, as quality and study design would be assessed in the Cochrane Review, thus reducing the potential number of studies eligible for inclusion.

Whilst the most common intervention type was therapy-based interventions, they were very diverse, meaning that few could be compared in a Cochrane Review. However, the next most common type of intervention was skill-based interventions, which were predominately focused on tooth brushing skills.

Based on the findings of the Scoping Review, it became clear that focusing on a population with intellectual disabilities and an oral hygiene skills-based intervention would be a worthwhile research question for the Cochrane Review. The choice of outcomes for the Cochrane Review was supported by those found in the scoping review but limited to those relevant to the research question: gingival health, dental caries, behaviour, attitude, self-efficacy and quality of life.

Chapter 4 Cochrane Review

4.1 Introduction

The Cochrane Review process began in June 2015 and will be published early in 2019. It is a regimented, thorough and slow process, with distinct stages, all of which must get editorial approval and be peer reviewed before publication. However, this arduous process ensures the quality of the review and is what makes a Cochrane Review stand apart from other systematic reviews. Table 12 outlines the time frame for the various stages of this review.

Table 12: Stages and time frames for the Cochrane Review

Stages of the Cochrane Review	Time frame
Discussion with Cochrane Oral Health Group (COHG) re focus of title	June 2015
Title Application Form submitted	October 2015
Title submission reviewed by COHG Editorial Team	January 2016
Title accepted	February 2016
Protocol drafting with team – 3 major iterations	April – Sept 2016
Draft Protocol submitted	September 2016
Protocol reviewed by COHG Editorial Team	November 2016
Protocol reviewed by three peer reviewers	December 2016
Feedback from Reviewers received	February 2017
Amended protocol resubmitted	March 2017
Final Protocol Published	April 2017
Review process • Searches of Databases and trial registers • Findings Screened by Title and Abstract • Sourcing articles and full text screening • Data extraction and Management • Assessment of Risk of Bias • Summary of findings Tables • Data Synthesis and Analysis • Assessment of quality of findings	April 2017 June 2017 July 2017 – Sept 2017 Sept 2017 – January 2018 Sept 2017 - January 2018 February 2018 March 2018 – October 2018 March 2018 – October 2018
Review drafting by team	November 2018
Draft Review submitted	December 2018
Review and feedback from by COHG Editorial Team	December 2018
Amendments made and resubmitted	January 2019
Review by three peer reviewers - pending	January 2019
Final Review Publication - pending	February / March 2019

The presentation of the Cochrane Review in this thesis follows the format outlined by Cochrane on the RevMan database. It starts here with the Background; the Abstract and Plain language summary will precede this section in the published review.

4.2 Background

4.2.1 Description of the condition

Oral hygiene is the process of cleaning the hard and soft tissues of the oral cavity (teeth, gums and tongue), fixed dental prostheses, oral appliances and dentures. Failure to perform regular and effective oral hygiene results in increased levels and varieties of the dental bacteria that make up dental plaque. Dental plaque has been shown to have a major role in the development of periodontal disease and dental caries, the two most common oral diseases (Axelsson 2004; Löe 2000)

Poor oral hygiene can affect a person's quality of life because of pain and discomfort when eating, poor self-esteem or sleep disturbance as a result of dental caries (Bonetti *et al* 2015). Evidence also exists linking levels of oral hygiene and periodontal status to diabetes, aspiration pneumonia and rheumatoid arthritis and evidence is emerging of a link with other systemic diseases and conditions such as cardiovascular disease, liver disease and obesity (Borgnakke 2015; Horwitz *et al* 2000).

People with an intellectual disability (ID) have a higher prevalence and greater severity of periodontal disease than the general population (Anders & Davis 2010; Campanaro *et al* 2014). The prevalence of dental caries in children with ID is similar to those of the same age in the general population (Whelton *et al* 2009). However, their oral health deteriorates at a faster rate as they move into adulthood. There are more untreated dental caries, more missing teeth and fewer restorations found in adults with ID than in the general population (Anders & Davis 2010; Catteau *et al* 2011; Crowley *et al* 2005; De Jongh *et al* 2008; Hennequin *et al* 2008; Morgan *et al* 2012; Oliveira *et al* 2013).

In the general population, brushing regularly with fluoridated toothpaste has been shown to play an important role in preventing dental caries and periodontal disease (Axelsson *et al* 2004; Broadbent *et al* 2011; Chestnutt *et al* 1998; Loe 1965; Loe 2000; Van der Weijden & Slot 2011; Zimmermann *et al* 2015). Whether this also applies to people with an ID is less clear, as many research studies on oral health exclude people with disabilities (Glassman & Miller 2009).

The removal of dental plaque may reduce the prevalence of periodontal disease and dental caries (Axelsson & Lindhe 1978; Axelsson *et al* 2004; Loe *et al* 1965; Loe 2000). However, the plaque must be removed thoroughly and regularly to achieve this effect. This health-related behaviour requires an individual to have the relevant capabilities, opportunities and motivation to perform the necessary routines (Broadbent *et al* 2011; Stewart *et al* 1996; Watt & Marinho 2005). For people with an ID, understanding the importance of removing dental plaque and developing the skills to do so requires more effort and achieving the standard of oral hygiene necessary for good oral health may require the support of others (Crowley *et al* 2005).

When considering the issues possibly influencing oral hygiene practices for the population with ID, it is important first to have a clear definition of ID; second, to consider the specific barriers to regular oral hygiene practices that might exist for people with ID; and third, to consider facilitators to promoting regular oral hygiene practices that might exist for people with ID.

4.2.2 Definition of intellectual disability

The most widely used current definition of disability comes from the World Health Organization's (WHO) International Classification of Functioning, Disability and Health (ICF), which incorporates the complex interactions between health conditions, environmental factors and personal factors. Regarding a person with an ID, this definition would consider how their personal factors, health condition and environment affect their lives (WHO 2001). Three elements are common for people with ID: a significant impairment of intelligence; a resultant significant reduction in adaptive

behaviour/social functioning; and the development of the condition before the age of 18, which persists throughout life (Schalock *et al* 2010).

The WHO has also developed an operational definition of ID, which focuses mostly on the functional elements of ID, in the International Statistical Classification of Diseases and Related Health Problems, 10th version (ICD-10) (WHO 2011) (Table 13). The ICF definition of disability is designed to complement the ICD-10 definition.

Table 13: The ICD-10 levels of ID

Mild intellectual disability (ICD10 F70) IQ range 50 to 69	"Ability to use speech in everyday situations; usually full independence in self-care; Difficulties in identification of this population arise, as those with borderline disability may not be in contact with service providers."
Moderate intellectual disability (ICD10 F71) IQ between 35 and 49	"Slow in comprehension; supervision of self-care, retarded motor skills."
Severe intellectual disability (ICD10 F72) IQ between 20 and 34:	"Marked impairment of motor skills; clinically significant damage to Central Nervous System (CNS)."
Profound intellectual disability (ICD10 F73) IQ less than 20	"Severely limited understanding; Immobility or restricted mobility; Incontinence; requires constant supervision; usually organic aetiology."

We acknowledge that other terms and definitions have been used to describe people with ID, both historically and currently. Whilst we acknowledge that the ICF is currently accepted as the most holistic definition of disability, we considered it unlikely to have been used in the studies that were potentially eligible for this review. We therefore planned to use the ICD-10 definition as the reference to determine the inclusion or exclusion of studies, as it provides a detailed nomenclature of diseases (Kurbasic *et al* 2008). In future updates to this review, the ICF definition of ID will have become more commonly used

in studies and will more accurately reflect the make-up of the population with disabilities. In addition, the ICF definitions are a better representation of populations requiring targeted services. However, the definition used (ICD-10) was felt by the team to accurately represent the population of interest in this review.

4.2.3 Barriers to oral hygiene practice for people with ID

Capability, opportunity and motivation are required to achieve adequate oral hygiene. Oral hygiene tasks are more difficult for people with ID; they may lack the manual dexterity or cognitive skills to carry out or understand the need for effective oral hygiene (Anders & Davis 2010; Loe 2000). There may be other associated conditions such as sensory impairment, anxiety or chronic medical or behavioural conditions that militate against carrying out oral hygiene effectively (Gordon *et al* 1998; Minihan *et al* 2014; Prangnell & Green 2008). In a 2003 Irish study, carers of people with ID identified some of the physical barriers caused by the disability such as an inability to rinse, difficulty getting a toothbrush into someone's mouth, an oversensitive mouth and a tendency to gag (Taylor-Dillon *et al* 2003). Nearly 10% of people with ID have dysphagia, which is associated with an increased tendency to choke or gag during toothbrushing (Chadwick *et al* 2014).

People with ID have been shown to have chronic, poor oral hygiene and to require more support with their oral hygiene. Carers for people with ID are often poorly trained (Crowley *et al* 2005). Oral hygiene is not seen as a priority for overstretched carers, particularly if it is not a personal priority (Kaye *et al* 2005; Rawlinson 2001). Lack of co-operation and challenging behaviour on the part of the person with ID may add to this burden.

4.2.4 Facilitators for oral hygiene practice for people with ID

Behaviour management techniques may improve the co-operation of individuals with ID when performing oral hygiene. Education may change carers behaviour and management techniques may improve the co-operation of individuals with ID when performing oral hygiene. Education may change carers' attitudes and improve their skills (Faulks & Hennequin 2000; Mac Giolla

Phadraig *et al* 2013). Management support and guidance, stakeholder engagement, policy documents and increased resources may improve levels of oral hygiene provision in community and institutionalised settings (Glassman & Miller 2003; Glassman & Miller 2006; Glassman & Miller 2009). Specially designed or adapted oral hygiene aids may improve the oral hygiene skills of people with ID or their carers (Dougall & Fiske 2008b).

4.3 Description of the intervention

The interventions evaluated in this Cochrane Review focused on two distinct groups: people with an ID and the carers supporting, assisting or providing oral hygiene care for them. The range of carers involved was expected to be diverse; formal and non-formal personal carers with and without relevant training, with and without dental qualifications and with and without psychology or related qualifications.

We defined oral hygiene in this review as the mechanical removal of plaque with either manual or electric toothbrushes, interdental aids or other mechanical aids. Interventions designed to impact on levels of oral hygiene tend to categorise outcomes as clinical, such as reductions in plaque levels, periodontal diseases or dental caries, and behavioural/psychological, such as changes in comprehension, self-efficacy or motivation (Cooper *et al* 2013; Cooper *et al* 2014). This review included interventions that assessed clinical or behavioural/psychological outcomes or both. We considered any intervention that may have impacted on the duration, frequency and/or effectiveness of oral hygiene practices.

4.3.1 How the intervention might work

Behavioural change models have been shown to relate to oral hygiene behaviour and to be effective when used in interventions for people with ID (Renz *et al* 2007; Willems *et al* 2017). In a systematic review, Newton & Asimakopoulou (2015) looked at using the constructs of changing behaviour rather than the models themselves and found this to be a more reliable way of predicting behavioural change. Michie and colleagues (2013) have identified a taxonomy of 93 behaviour change techniques (BCTs) that provides a

standardisation of the terminology used. In order to make the taxonomy of BCTs more accessible for clinicians, the BCTs can be grouped into 16 labelled clusters (Cane *et al* 2015; Michie *et al* 2011; Michie *et al* 2013). These labels provide a means to identifying the elements in an intervention that are likely to improve adherence and make routines easier to establish in relation to oral hygiene, even if they are not formally identified as BCTs. To further assist the analysis of interventions, Michie developed the 'Behavioural Change Wheel', which characterises the elements at three levels: a behaviour system at the hub, encircled by intervention functions and then by policy categories.

For this review, the interventions were considered under the "behaviour system" elements of Capability (which includes knowledge and skills), Opportunity (both physical and social) and Motivation (both automatic and reflexive), as outlined by Michie as the COM-B framework for understanding behaviour and behavioural change (Michie *et al* 2011).

Capability-based interventions, defined as the psychological and physical capacity to engage in the activity concerned, might be a knowledge-based intervention in relation to the comprehension and reasoning around why oral hygiene is required or a skills-based intervention that trains a person with ID in the physical skills of toothbrushing or that trains carers how to brush the teeth of a person with ID.

Opportunity-based interventions, which include both physical and social opportunities, should consider the elements beyond the individual, that is contexts such as the variety of settings in which the intervention is delivered or undertaken, for example, home, school, dental clinic, day care or residential care setting or the influences of the families or carers' attitudes regarding oral hygiene practices.

Motivation-based interventions, which include both automatic and reflective processes, might result in conscious decision-making where a goal or reward is identified to instigate or perpetuate a daily oral hygiene routine.

An intervention may change one or more of these elements resulting in the desired behaviour of performing regular oral hygiene.

4.4 Why it is important to do this review

The difficulties of providing dental care for those with disabilities have been acknowledged (Al Kindi & Nunn 2016). Access to care is known to be more difficult; resources are limited for parents, carers and healthcare professionals (Tiller *et al* 2001; Kaye *et al* 2005; Prabhu *et al* 2010; Sagheri *et al* 2013). The consequences of oral diseases for people with ID are more profound. Interventions designed to suit people with disabilities must show benefit (WHO 2011). Whilst an increasing number of good-quality systematic reviews of oral health interventions designed for the general population have been produced (Agnihotry *et al* 2016), Cochrane has not specifically reviewed the effectiveness of oral health interventions focused on people with disabilities (Cochrane Oral Health 2018).

The scoping review of interventions aimed at the wider population of people with disabilities has identified those with ID as being the most common target subpopulation (Waldron *et al* 2016). Oral hygiene status is one of the most common targets of oral health intervention for those with disabilities. This systematic review of oral hygiene interventions for people with ID allows us to draw conclusions about the effectiveness of these types of interventions. Furthermore, the review may increase dental professionals' understanding of behaviour change principles, inform the training of the dental team and have an impact on the design and implementation of future oral health interventions for this population.

Cochrane Oral Health undertook an extensive prioritisation exercise in 2014 to identify a core portfolio of titles that were the most clinically important ones to maintain on the Cochrane Library (Worthington *et al* 2015). This exercise identified prevention and maintenance as priorities, which included oral hygiene adherence and caries prevention. Whilst not specifically identifying people with disabilities, the exercise also prioritised oral health promotion, specifically in institutionalised settings (Worthington *et al* 2015).

Objectives

To assess the effects (benefits and harms) of oral hygiene interventions for people with intellectual disabilities (ID).

4.5 Methods

4.5.1 Criteria for considering studies for this review

Types of studies

We included RCTs, including cluster-RCTs, evaluating interventions targeted at people with ID and groups such as parents, carers and other healthcare professionals caring for people with ID. As we anticipated that the number of RCTs was likely to be low, we included some types of NRS: non-randomised controlled trials, controlled before-after studies, interrupted time series studies and repeated measures studies.

Interrupted time series, where multiple data points are collected before and after the intervention and the intervention effect is measured against the pre-intervention trend and repeated measure studies, where these measures are taken on the same individuals at each time point, will only be included if they have a clearly defined point in time when the intervention occurred and at least three data points before and three after the intervention.

We analysed RCTs and NRS separately, and present RCT findings as the key evidence. We applied no language restrictions.

Types of participants

People with an ID, living either at home, in day care, in hospital or in a residential care setting. We considered all intellectual disabilities as defined by the ICD-10 (WHO 2011).

Types of interventions

We considered all oral hygiene interventions, including but not limited to the following components:

- Oral hygiene-related knowledge-based interventions for participants or their carers, e.g. education in the care of the teeth and gums, diseases of the teeth and gums and prevention of diseases of the teeth and gums.
- Oral hygiene-related behavioural interventions for participants or their carers that result in a behavioural change that can be observed and is replicable, e.g. goal setting or positive reinforcement.

- Oral hygiene-related skills training for participants, e.g. interventions related to toothbrushing skills or other oral hygiene routines.
- Oral hygiene-related skills training for carers, e.g. interventions related to assessing toothbrushing skills or assisting participants with toothbrushing or other oral hygiene routines or providing oral hygiene care to participants.

To be eligible, controlled studies must have included a comparison with at least one of the following: usual care, no care or a similar alternative intervention. Examples of studies comparing similar alternative interventions might be a study comparing education provided in different formats, for example, face to face versus an information leaflet or a study comparing oral hygiene care provided after a training session versus oral hygiene care provided without training.

Types of outcome measures

Outcome measures included both behavioural and clinical measures.

We based the behavioural outcomes measured on the Capabilities, Opportunities and Motivation elements of COM-B framework and included indicators such as improved toothbrushing skills, changes in knowledge of participants and carers with regard to oral health, and increased episodes and duration of toothbrushing. We accepted any reasonable instrument used in the included studies such as questionnaires, interviews, observational outcomes, video recordings, diaries, self-reported outcomes or observational measures such as measuring toothpaste weight to check adherence with toothbrushing.

The clinical outcomes measured included indicators such as the prevalence/incidence of dental plaque, prevalence/incidence of periodontal disease (gum disease) and prevalence/incidence of dental caries. We used only published or validated assessments of the clinical outcomes or a comparable measure, for example, dental plaque measured using the Plaque Index, as outlined in Silness 1964 or Quigley and Hein's Plaque Index as modified by Turesky and colleagues (Turesky *et al* 1970). Gingivitis measured using an index such as the Gingival Index, as outlined in Löe 1963 and Löe 1967 or the Modified Gingival Index (Lobene *et al* 1986). Dental caries was to be measured

using an index such as the Decayed, Missing or Filled Teeth/Surface Index (dmft/s; DMFT/s) (Petersen & Baez 2013).

We considered all time frames for the delivery and follow-up of the intervention. The follow-up time frames were grouped for analysis into short, medium and long term, for all outcomes being measured. A systematic review of the effects of oral hygiene on chronic periodontitis in the general population excluded studies lasting less than nine months, in order to differentiate between gingivitis and periodontitis (Hujoel *et al* 2005). The three RCTs included in that review were followed up for over three years. Mombelli (1998) reflected on the limitations of longitudinal studies of periodontal disease when considering the issues of measurement error in periodontal probing and the impact of concepts such as bursts of disease activity versus a continuous disease process, in relation to the progression of periodontal disease. Mombelli (1998) concluded that studies of 20 years or more in length would be required to achieve sufficient confidence in the results. A recent systematic review on the effects of toothbrushing frequency on the prevention of dental caries showed that the eligible interventions were all followed up for between 11 months and 15 years (Kumar *et al* 2016).

The division of time frames was guided by a review of the literature on the effects of oral hygiene interventions on the oral health of people with ID and the knowledge, behaviour, attitude and self-efficacy of the people with ID and/or their carers:

Short-term follow-up (< six weeks) may allow for some clinically visible signs of early changes in gingival health to become apparent, permitting the impact of possible differences in the microbial load, immune responses and environmental factors in this population group to manifest in clinically observable signs (Garmyn *et al* 1998). It may also be useful to observe the clinical changes resulting from an intervention without confounding the result with other variables, e.g. the effect on oral hygiene following skills training without the variable of adherence to oral hygiene practices over time (Egelberg & Claffey 1994). The early, clinically visible signs would include changes in

plaque prevalence/incidence and changes in gingival bleeding or inflammation (Löe *et al* 1965).

Medium-term follow-up (six weeks to 12 months) may allow for some additional signs of changes in the gingival health to be recorded compared to the baseline status, depending on the success of the intervention, such as gingival pocket probing depths, as well as consistent changes in knowledge and behaviour.

Long-term follow-up (> 12 months) would allow for some assessment of those measures that require a longer time frame to show change, such as dental caries prevalence/incidence or clinical attachment loss or bone loss in relation to gingival disease, as well as continued follow-up on all other assessments. Long-term studies would also allow for follow-up on any short- or medium-term success regarding changes in oral hygiene behaviour that is crucial to maintaining oral health (Schou 1998). It must be accepted that any findings from these long-term interventions will be confounded by factors outside the study setting.

Primary outcomes

- Gingival health of people with an ID, e.g. plaque, gingival inflammation, calculus.

Secondary outcomes

- Oral hygiene skills acquisition by people with an ID or the acquisition of skills by their carers in providing or supporting oral hygiene care.
- Oral hygiene knowledge acquisition by people with an ID or the acquisition of knowledge by their carers with regard to oral hygiene.
- Changes in behaviour, attitude and self-efficacy of people with an ID or their carers.
- Quality of life changes or long-term, patient-centred outcomes for people with an ID (e.g. tooth retention).
- Dental caries levels of people with an ID.
- Any unintended (positive or negative) effects of the interventions.

Outcome measures relating to clinical outcomes must have been published or validated or comparable to measurements such as dental plaque levels (i.e. Plaque Score), gingival health (i.e. Gingival Index) or dental caries prevalence/incidence (i.e. DMFT/S). Behavioural outcome assessments with regard to changes in capability, opportunity or motivation of the participant or carer for oral hygiene practices included any reasonable instrument used in the included studies.

4.5.2 Search methods for identification of studies

Electronic searches

Cochrane Oral Health's Information Specialist conducted systematic searches in the following databases for randomised controlled trials and controlled clinical trials without language or publication status restrictions:

- Cochrane Oral Health's Trials Register (searched 4 February 2019);
- Cochrane Central Register of Controlled Trials (CENTRAL; in the Cochrane Register of Studies searched 4 February 2019);
- MEDLINE Ovid (1946 to 4 February 2019);
- Embase Ovid (12 October 2016 to 4 February 2019);
- PsycINFO Ovid (1806 to 4 February 2019).

Details of the search strategies can be found in Appendix 1.

Subject strategies were modelled on the search strategy designed for MEDLINE Ovid. Due to the Cochrane Centralised Search Project to identify all clinical trials in the database and add them to CENTRAL, only the most recent months of the Embase database were searched. See the searching page on the Cochrane Oral Health website for more information. No other restrictions were placed on the date of publication when searching the electronic databases.

Searching other resources

The following trial registries were searched for ongoing studies (see details of search terms used in Appendix 1):

- US National Institutes of Health Ongoing Trials Register ClinicalTrials.gov (clinicaltrials.gov/);
- World Health Organization International Clinical Trials Registry Platform (apps.who.int/trialsearch).

We also hand searched the specialist conference abstracts from the International Association of Disability and Oral Health (2006 to 2016). Conference abstracts prior to 2006 were not published and were not available from other sources.

We checked the bibliographies of included studies and any relevant systematic reviews identified for further references to relevant trials (Horsley 2011).

We considered any unpublished studies discovered using the search methods described above if they met the inclusion criteria for this review.

We considered adverse effects described in included studies only.

4.5.3 Data collection and analysis

Selection of studies

Five review authors independently examined the studies identified by the searches and ensured that each study was examined by at least two review authors, firstly by the titles and abstracts and then by obtaining and reading the full text, if the study was considered potentially relevant. We used specific inclusion criteria in our decision-making including the participants and interventions as outlined below. A sixth review author was the arbiter.

Inclusion criteria:

- Participants must have an intellectual disability as defined by the ICD-10 or be a carer of a person with an intellectual disability.
- The intervention must relate to oral hygiene.
- Complex interventions with an oral hygiene element were considered if the outcome from the oral hygiene component could be appraised separately.

Details such as keywords, authors, publication and institutions were available to the review authors. Any disagreements were discussed by the reviewing authors and the arbiter. We recorded justifications for all decisions. When we identified multiple reports of the same study, we linked these together in so

far as this was possible. We contacted study authors to clarify details regarding eligibility where necessary. We attempted to identify all relevant studies irrespective of language. Relevant non-English language papers were translated. Any studies identified by the searches, involving any of the review authors, were examined by other review authors.

Data extraction and management

Two review authors independently extracted data for each study using a specially designed data collection sheet. We extracted data under the following headings: profile of the participants, number of participants, settings, details of interventions including details of the carers and/or personnel delivering or supporting the intervention and any behavioural change techniques identified within the interventions, outcomes measured, duration of intervention and follow-up and study design. We designed a template for the 'Characteristics of studies' tables, based on the methods outlined in the protocol and headings used in the data collection sheet, to highlight the important elements of the studies (Appendix 1).

Assessment of risk of bias in included studies

Two review authors independently assessed the risk of bias for each study, discussing any disagreements with the arbiter. We recorded justifications for decisions. To avoid any conflict of interest, we excluded team members who were authors of included studies from the 'Risk of bias' assessment of those studies and alternative review authors undertook this assessment.

We considered bias for RCTs and NRCT for the following domains: selection bias, performance bias, detection bias, attrition bias and reporting bias as per the *Cochrane Handbook for Systematic Reviews of Interventions* (Higgins *et al* 2011a). We considered bias for cluster-RCTs under the two different units of measurement, the cluster and the participant, if provided and under the same domains as for RCTs (Campbell *et al* 2012).

We considered bias for non-controlled trials using the Effective Practice and Organisation of Care (EPOC) criteria for assessing risk of bias (EPOC 2017; Sterne *et al* 2016) under the following headings:

- Was the intervention independent of other changes?
- Was the shape of the intervention effect pre-specified?
- Was the intervention unlikely to affect data collection?
- Was knowledge of the allocated interventions adequately prevented during the study?
- Were incomplete outcome data adequately addressed?
- Was the study free from selective outcome reporting?

We supplemented this with information on participant and study characteristics where appropriate.

We completed a 'Risk of bias' table for each included study and rated each study as either high, low risk or unclear risk of bias for the separate domains and presented the results graphically (Figure 4, page 87; Figure 5, page 92). All domains were regarded as equally important in this review. The overall risk of bias within a study was determined across domains depending on the number of low, unclear and high ratings: if there was at least one rating of high risk of bias for the study, it was rated overall as high risk of bias; if all ratings were low risk of bias, the study was rated low overall; if there was a mix of low and unclear ratings, the study was rated overall as being at unclear risk of bias.

Measures of treatment effect

For continuous data (e.g. plaque levels, self-efficacy scale), we planned to use the mean difference (MD) when the same scale was used or standardised mean difference (SMD) if more than one scale was used to measure the same outcome, along with the 95% confidence interval (CI). We planned to use the risk ratio (RR) and its 95% CI for dichotomous data, for example, toothbrushing undertaken or not.

Unit of analysis issues

We considered the unit of analysis with regard to whether the intervention was targeted at individual or group level, whether there was more than one element to the intervention and whether there were different time points measured in the intervention. When possible, we planned to adjust for clustering if

participants were allocated to the same intervention in groups (e.g. family members or care homes) for example, in cluster-RCTs. To account for the effect of clustering, we planned to estimate inflated standard errors (SEs) (Deeks *et al* 2011).

With regard to RCTs and other comparative studies, we considered adjustments to avoid multiple use of participants in multi-arm or cross-over studies. If multiple arms were to be included in the meta-analysis in such a way that one intervention arm was to be included in more than one treatment comparison, we planned to divide the number of participants (in that arm) and the number of events by the total number of treatment comparisons. This approach retains original information but reduces the precision of the pooled estimates.

With regard to cross-over trials, data from these needs to be reported in a specific way to allow their inclusion in meta-analyses (e.g. showing the data from each treatment period to allow calculation of the standard error of the mean difference or data to approximate the correlation between results from different treatment periods). However, the necessary data were not available from the cross-over trials that we identified.

Dealing with missing data

We made all reasonable efforts to contact authors for details and reasons for any missing outcome data (Young & Hopewell 2011). If data were missing and no information was available in the paper or was not obtainable by contacting the authors, we planned to assume, for dichotomous data, that drop out was due to the intervention being unsuccessful. No dichotomous data were reported in this review. We planned to perform sensitivity analyses to assess the effects of these assumptions, for example, missing data for intervention studies could have been dealt with by using intention-to-treat numbers. For continuous data, we planned to use this information if we were able to calculate the data from other data provided, for example, calculating standard deviation (SD) from standard error (SE) and P values (Higgins *et al* 2011b).

Assessment of heterogeneity

We planned to assess levels of statistical heterogeneity by observing the confidence interval within studies and, to a lesser extent, statistically by use of the Chi^2 test to determine whether observed differences in results are compatible with chance alone. Interpretation of the Chi^2 would take into consideration the sample size and number of studies included in the meta-analysis, detailed under the data synthesis section.

We planned to quantify heterogeneity with the I^2 statistic, using the following guide. Some level of heterogeneity was expected (Higgins *et al* 2011a).

- 0% to 40%: might not be important
- 30% to 60%: may represent moderate heterogeneity
- 50% to 90%: may represent substantial heterogeneity
- 75% to 100%: very substantial ("considerable") heterogeneity.

Assessment of reporting biases

Thorough searching of multiple databases should limit general reporting biases. We kept reporting biases due to such things as time lag bias (Hopewell *et al* 2007), selective outcome reporting (Dwan *et al* 2014) and publication bias (Hopewell *et al* 2009), to a minimum by inclusion of prospective trial registers; if we identified at least ten studies, we planned to use funnel plots to explore publication bias and other biases related to study size. We made every effort to contact authors for results of outcomes included in their study objectives but not reported.

Data synthesis

We undertook meta-analyses only when studies were sufficiently homogeneous in their participants, interventions and outcomes; otherwise we present a narrative of individual study findings. We planned to combine RRs for dichotomous data and MDs or SMDs for continuous data, using random-effects models if at least four studies were included in a meta-analysis. We used fixed-effect models when there were fewer than four studies and when heterogeneity was reasonably low.

Subgroup analysis and investigation of heterogeneity

We planned to consider subgroup analysis for topics such as the following if sufficient data were available (Bender *et al* 2008).

- Level of intellectual disability
- With versus without support of dental professionals
- Non-formal carers (i.e. parents or siblings) versus formal carers
- Setting of the intervention (i.e. home-based versus residential / institutional / school-based interventions)
- Behavioural change technique used based on the COM-B framework
- Short-term interventions versus long-term interventions

For most of the subgroups, the amount of data was too small to undertake any purposeful analysis.

Sensitivity analysis

Providing there were sufficient included studies, we planned to undertake sensitivity analyses based on risk of bias; however, there were insufficient studies for a sensitivity analysis of this kind. We undertook a post hoc analysis excluding studies that included participants with IQ above 70.

Summarising findings and assessing the quality of the evidence

We created 'Summary of findings' tables for the nine comparisons identified; six had data that could be included in a meta-analysis. The other three comparisons were summarised narratively (Appendix 4).

1. Special manual toothbrushes compared to conventional manual toothbrushes for people with intellectual disabilities
2. Electric toothbrushes compared to manual toothbrushes for people with intellectual disabilities
3. Oral hygiene care training compared to no oral hygiene care training or a placebo training session for carers of people with intellectual disabilities
4. Oral hygiene training compared to no oral hygiene training for people with intellectual disabilities
5. One-, three- and six-monthly dental recall intervals compared to no dental recall for people with intellectual disabilities

6. Use of individual clinical photographs as a toothbrushing motivator compared to no motivator for people with intellectual disabilities
7. Daily tooth brushing compared to twice weekly or once weekly tooth brushing for people with intellectual disabilities
8. Use of a plaque disclosing agent compared to no plaque disclosing agent for people with intellectual disabilities
9. Individualised oral care plan compared to no oral care plan for people with intellectual disabilities

We summarised the findings for the outcomes measured (plaque, gingival inflammation, gingival bleeding, calculus, oral health, knowledge, behaviour, attitude and self-efficacy) over the three-time frames (short, medium and long). We also used GRADEpro software to assess the certainty of the evidence under the following headings: study design (risk of bias); inconsistency; indirectness; imprecision; publication bias; large effect; plausible confounding; dose response gradient (Schünemann *et al* 2013). We categorised the quality of the body of evidence for each of the main outcomes for each comparison as high, moderate, low or very low.

4.6 Results

4.6.1 Description of studies

The results of this Cochrane Review are outlined under the following headings and sub-headings, as they appear in RevMan:

Results of the search

Included studies: characteristics of the methodologies; characteristics of the participants; characteristics of the interventions; characteristics of the outcomes; COM-B system characteristics; stakeholder involvement; funding; adverse effects

Excluded studies: reasons for exclusion

Risk of bias in included studies: allocation (selection bias); blinding (performance bias and detection bias); incomplete outcome data (attrition bias); selective reporting (reporting bias); other potential sources of bias. Risk of bias for Interrupted Time Series and Repeat Measure studies

Effects of interventions: Comparisons 1 - 9

4.6.2 Results of the search

The searches from the databases resulted in 3805 records and an additional 41 records were identified from other sources. After removal of duplicates, review authors screened 3020 records by title and abstract using Covidence software (Covidence 2018) of which 2794 were judged to be irrelevant and rejected. Following this process, we attempted to source 226 full-text records for assessment for eligibility against the inclusion and exclusion criteria outlined in the protocol. We rejected 153 records. We excluded 23 records (22 studies) for reasons listed in Characteristics of excluded studies; three studies are awaiting classification (see Characteristics of studies awaiting classification), and one is ongoing (see Characteristics of ongoing studies). Therefore, a total of 34 studies (46 records) met the inclusion criteria for this review (Figure 3).

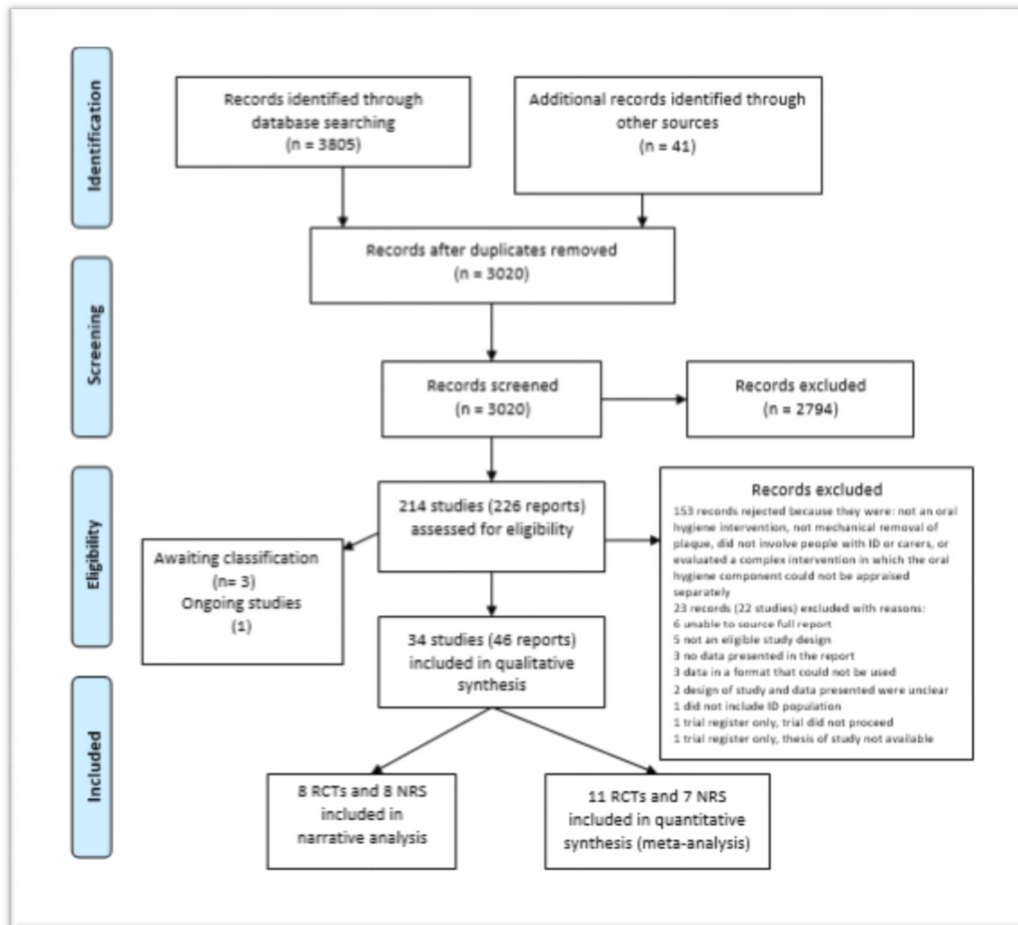


Figure 3: Prisma Flow Chart for the Cochrane Review

Data extraction was first piloted by three of the review authors on a small number of records and some clarifications to the criteria were made following a team discussion. In relation to the definition of oral hygiene, we made the following clarification: Interventions focusing on the chemical removal of plaque alone will be excluded. The use of a chemical agent, which is used on a toothbrush and arguably removes plaque both mechanically and chemically, is complex, as the measurement of the resulting reduction of plaque cannot be assigned to either solely the mechanical or chemical action. These studies should be excluded, except for those studies using (a) a conventional over the counter toothpaste normally used by the participants or (b) using a non-conventional chemical agent for both the intervention and control in studies comparing the mechanical removal of plaque. For example, a study comparing

a manual toothbrush with an electric toothbrush where both are used with a non-conventional toothpaste such as Chlorohexidine or extra strength fluoride toothpaste. Screening and extraction were undertaken by at least two review authors independently; three review authors were involved in the screening of titles and abstracts (CW, JN, CMGP), five review authors were involved in the extraction process (CW, JN, CMGP, SG, CC).

4.6.3 Included studies

We designed a template for the 'Characteristics of studies' tables, based on the methods outlined in the protocol, to highlight the important elements of the studies (Appendix 3). The study details are summarised here under the headings used in this template and the details of each included study are available in the Characteristics of included studies tables (Appendix 5).

Characteristics of the methodologies:

Design

Thirty-four studies met the inclusion criteria for this review, 19 RCTs and 15 NRSs. RCTs and NRSs are reported separately (11 RCTS and 7 NRS were included in the meta-analyses; 8 RCTs and 8 NRS were reported narratively).

Of the 19 RCTs, fifteen studies were conventional RCTs (Albino 1979; Bickley 1990; Bildt 2010; Christen 2007; Dôgan 2004; Ferozali 2007; Gonzalez 2013; Kaschke 2005; Kelner 1963; Lange 1985; Olmos 2016; Phlypo 2016; Sauvetre 1995; Shaw 1983; Swallow 1969) and four were cluster RCTs (Carr 1997; Garcia-Carrillo 2016; Mac Giolla Phadraig 2015; Shaw 1991). Four of the RCTs were cross-over trials: one trial had two arms (Shaw 1983) and the others had three arms (Christen 2007; Dôgan 2004; Kaschke 2005).

Of the NRS, nine studies were non-randomised controlled trials (NRCT) (Altabet 2003; Bratel 1991; Gertenrich 1967a; Gertenrich 1967b; Gertenrich 1967c; Gertenrich 1967d; Lange 2000; Teitelbaum 2009; Williams 1988), three were interrupted time series (ITS) studies (Glassman 2006; Jarman 1983; Kissel 1983) and three were repeat measure (RM) studies (Abramson 1972; Bouter 1979; Snell 1989).

Outcomes

The clinical outcomes assessed for the participants in the RCTs were: plaque, oral hygiene status (subjective), gingival inflammation, gingival bleeding, calculus, oral assessment, total bacterial count and potential pathogenic bacteria count. The behavioural outcomes assessed for the participants and/or the carers were knowledge, behaviour, attitude and self-efficacy.

The outcomes assessed for the participants in the NRS were: plaque, oral hygiene status, gingival inflammation, gingival bleeding, gingival pocketing, calculus, self-efficacy (steps in a toothbrushing routine), duration of the toothbrushing episode, individually tailored expected toothbrushing performance and level of "self-initiation". The outcomes assessed for the carers in the NRS were: need for verbal instruction and physical guidance, carer presence during toothbrushing by a person with ID and carer use of verbal instruction, physical guidance and rewards.

An extraction form template for each of the study designs was created (Appendix 6), which was then tailored to each study and used to record the statistical data for that study, the outcomes assessed, the indices used and any additional data in relation to the results of the intervention being reported. The extraction form templates and an example of a completed extraction form (Appendix 7) are provided in the Appendices.

Date, duration and size of studies

The 19 RCTs were reported between 1963 and 2016. One RCT included long-term follow-up, (24 months) (Shaw 1991); 8 included medium-term follow-up (> six weeks to < 12 months) (Bickley 1990; Bildt 2010; Carr 1997; Ferozali 2007; Garcia-Carrillo 2016; Kelner 1963; Mac Giolla Phadraig 2015; Phlypo 2016); and 10 had only short-term follow-up (< six weeks) (Albino 1979; Christen 2007; Dôgan 2004; Gonzalez 2013; Kaschke 2005; Lange 1985; Olmos 2016; Sauvetre 1995; Shaw 1983; Swallow 1969).

The 15 NRS were reported between 1967 and 2009. Two NRS included a long-term follow-up (16 to 19 months) (Bratel 1991; Snell 1989), 10 included a

medium-term follow-up (Altabet 2003; Bouter 1979; Gertenrich 1967a; Gertenrich 1967b; Gertenrich 1967c; Gertenrich 1967d; Glassman 2006; Jarman 1983; Kissel 1983; Williams 1988) and two had only short-term follow-up (Lange 2000; Teitelbaum 2009). The duration of the follow-up was unclear for one NRS (Abramson 1972).

The number of participants in the studies is based on the number of participants recorded at baseline; some studies reported the intention to treat numbers and some did not. The number of participants in the RCTs ranged from 18 to 329. Nine RCTs had less than 50 participants, six RCTs had between 50 and 100 participants and four RCTs has more than 100 participants (Kelner 1963; Mac Giolla Phadraig 2015; Olmos 2016; Shaw 1991).

Most studies evaluated interventions aimed specifically at those with intellectual disabilities. Four of the RCTs included carers as participants, one assessed outcomes for the carers only (Gonzalez 2013); three assessed outcomes for both the people with ID and the carers (Mac Giolla Phadraig 2015; Olmos 2016; Phlypo 2016), but one did not report the number of carers involved (Olmos 2016). The number of participants in the NRS ranged from 3 to 79. Four NRS had less than 20 participants, nine NRS had between 20 and 50 participants and two NRS had more than 50 participants (Gertenrich 1967a; Altabet 2003). Two NRS assessed outcomes for both people with ID and carers (Glassman 2006; Kissel 1983); one NRS involved carers in the study but only assessed outcomes for the people with ID (Lange 2000).

Location, language and setting

The studies were carried out in 10 different countries: Belgium (two RCTs), Brazil (one NRS), Germany (three RCTs), Ireland (one RCT), Spain (one RCT), Sweden (one NRS), the Netherlands (one RCT and one NRS), Turkey (one RCT), the United Kingdom (four RCTs) and the United States of America (six RCTs and 12 NRS). Three of the included studies were in German (Christen 2007; Kaschke 2005; Olmos 2016) and one in Dutch (Bildt 2010).

The settings of the RCTs were residential (11 studies), school (four studies), day centre (three studies), home (one study) or mixed (two studies). One

setting was 'unclear'. The settings of the NRS were residential (13 studies), home (one study) or mixed (one study).

Ethical approval and consent

Twenty-two studies did not report having ethical approval; 10 RCTs and one NRS reported having received some form of ethical approval; one RCT was unclear on the issue. The participants or guardians gave consent in 13 of the RCTs and three of the NRS; four RCTs and 12 NRS did not report on consent; two RCTs were unclear.

Characteristics of the participants:

Participants with intellectual disabilities

In relation to people with ID, the studies included 1795 participants at baseline. In some studies, the difference between intention-to-treat numbers and those actually included at baseline was not explained. A total of 92 participants were reported as lost to follow-up; however, this number should be treated with caution as it is possible that those participants included at baseline were the more compliant participants and other potential participants were lost before baseline data could be collected.

Most studies had both male and female participants (25); one study had only male participants (Abramson 1972); five studies gave no details of the sex of participants; three studies were unclear.

The descriptions of the participants' level of intellectual disability used in the studies was converted to the ICD-10 descriptors: mild, moderate, severe and profound. The level of ID was unclear in seven of the RCTs (Bickley 1990; Carr 1997; Gonzalez 2013; Kelner 1963; Olmos 2016; Sauvetre 1995; Shaw 1991) and one NRS (Gertenrich 1967a). One RCT (Dôgan 2004) and one NRS (Gertenrich 1967d) had participants with a mild ID; three NRS had participants with a moderate ID (Bouter 1979; Bratel 1991; Teitelbaum 2009); one RCT (Shaw 1983) and one NRS (Jarman 1983) had participants with a severe ID; and one RCT (Ferozali 2007) and one NRS (Williams 1988) had participants

with a profound ID. All the other studies had participants with mixed levels of ID (17).

Two studies reported including some participants with an IQ above 70 (ranging as high as 87 and 94, respectively) (Garcia-Carrillo 2016; Gertenrich 1967c). We undertook a sensitivity analysis to determine the effect on the effect size of the outcomes of excluding these studies. No relevant effect was noticed.

Three RCTs (Albino 1979; Dôgan 2004; Shaw 1983) and six NRS (Abramson 1972; Gertenrich 1967b; Gertenrich 1967d; Kissel 1983; Snell 1989; Teitelbaum 2009) had participants who were aged less than 18 years; 15 studies had participants who were aged 18 years or older, with an age range of 18 to 64 years. Seven studies recruited both children and adults and four studies did not report the age profile.

Fifteen studies reported that participants had comorbidities: nine RCTs (Albino 1979; Bildt 2010; Carr 1997; Christen 2007; Ferozali 2007; Garcia-Carrillo 2016; Kaschke 2005; Shaw 1983; Shaw 1991) and six NRS (Gertenrich 1967a; Gertenrich 1967b; Gertenrich 1967c; Glassman 2006; Jarman 1983; Snell 1989). One study reported that participants were excluded if they had any systemic disease; 17 studies did not report on existing comorbidities; and one study was unclear. Of the studies reporting comorbidities, the most frequent conditions reported were cerebral palsy (five), epilepsy (four), Down syndrome (three), visual impairment (three) and hearing impairment (two). Other conditions included; anxiety disorder, asthma, autism, chronic bronchitis, depression, diabetes, emesis, enteral feeding, gastroesophageal reflux disease, heart defect, Hepatitis C, hiatal hernia, Hunter syndrome, hydrocephaly, hypertension, hypothyroidism, muscular dystrophy, ocular tension, paranoid schizophrenia, PKU disorder, Prader-Willi syndrome, speech difficulties, swallowing problems, Tay-Sach disorder, thyroid problems, Turner syndrome, along with general terms to describe physical disabilities.

Participants who were carers

There were 17 studies that actively involved the carers of the people with ID in the delivery of the intervention, of these only seven involved the carers as

participants; six of these provided details of the number of carers; three RCTs (Gonzalez 2013; Mac Giolla Phadraig 2015; Phlypo 2016) and three NRS (Glassman 2006; Kissel 1983; Lange 2000). A total of 354 carers provided baseline data; a total of 92 were lost to follow-up.

Characteristics of the interventions:

Interventions involving electric and manual toothbrushes

Thirteen studies compared electric toothbrushes to manual toothbrushes, eight of these studies were RCTs. Four of the RCTs were cross-over trials, one with two arms (Shaw 1983); the other three studies had three arms; the third arm was a special manual toothbrush (Christen 2007; Dôgan 2004; Kaschke 2005). The other four RCTs were Carr 1997; Garcia-Carrillo 2016; Kelner 1963 and Swallow 1969. The five NRS were Bratel 1991; Gertenrich 1967a; Gertenrich 1967b; Gertenrich 1967c; Gertenrich 1967d.

As well as the three, three-armed, cross-over trials, three other RCTs compared a special manual toothbrush with a conventional toothbrush (Bildt 2010; Ferozali 2007; Sauvetre 1995). Two NRS compared a special manual toothbrush with a conventional toothbrush (Bratel 1991; Williams 1988).

The studies comparing electric toothbrushes to manual toothbrushes compared a variety of electric toothbrushes: Oral B Braun (3D) (Bratel 1991; Dôgan 2004), Teledyne Waterpik Sonic Speed (Kaschke 2005), Panasonic Dentacare-Sonodent (Christen 2007), Interplak (Carr 1997), Phillips Sonicare EasyClean (Garcia-Carrillo 2016), Oral B Arcuate Action (Gertenrich 1967a; Gertenrich 1967b; Gertenrich 1967c; Gertenrich 1967d) and Broxodent (Kelner 1963). Shaw 1983, did not identify the electric toothbrush used.

The studies comparing special manual toothbrushes also used a variety of toothbrushes: Collis Curve (Williams 1988), Superbrush (Bildt 2010; Christen 2007; Dôgan 2004; Kaschke 2005; Sauvetre 1995) and a single-use suction toothbrush (Ferozali 2007).

Some studies distinguished between "self-brushers", "assisted brushers" and "carer brushed" or supervised or unsupervised brushing. Some studies specified the timing and duration of the brushing, while others did not.

Interventions involving training of carers and people with ID

Four RCTs (Gonzalez 2013; Mac Giolla Phadraig 2015; Olmos 2016; Phlypo 2016) and three NRS (Glassman 2006; Kissel 1983; Lange 2000) focused on the training of carers to improve the oral health of the people with ID for whom they cared. The description of the training provided lacked detail, those that indicated the duration of the training delivered it in one session, which varied in length from 90 minutes (Gonzalez 2013), to three hours (Glassman 2006), to a full day of training (Mac Giolla Phadraig 2015). Some interventions provided training manuals for the carers (Mac Giolla Phadraig 2015; Phlypo 2016) while others provided continual feedback based on observation (Glassman 2006; Lange 2000), video recordings (Kissel 1983) or monitoring of daily plaque scores (Lange 2000). Some interventions included practical skills training as well as theory, which was delivered either as role play (Mac Giolla Phadraig 2015), demonstrated on one of the participants (Lange 2000) or individualised for each carer and the people with ID for whom they cared (Kissel 1983). The timing of the evaluation of the intervention varied: immediately after the delivery of the training (Gonzalez 2013), four weeks (Olmos 2016), eight weeks (Phlypo 2016), 100 days (Kissel 1983), 156 days (Glassman 2006) and 11 months (Mac Giolla Phadraig 2015). The training was delivered mainly by dental professionals except for one that was delivered by a psychologist (Kissel 1983).

Two RCTs (Albino 1979 and Lange 1985) and four NRS (Abramson 1972; Bouter 1979; Jarman 1983; Snell 1989) focused on the training of people with ID to improve their oral health. The interventions all focused on training in the practical skills of toothbrushing or in the participants partial participation in toothbrushing skills (Snell 1989) with only one providing some theory in relation to the concepts of oral health (Albino 1979). The participants level of ID spanned the entire range: mild/moderate (Lange 1985), moderate (Bouter 1979), moderate/severe (Abramson 1972; Albino 1979), severe (Jarman 1983) and severe/profound (Snell 1989). The Snell 1989 study was the only

study that involved the participants in the partial participation of toothbrushing: the carers performed the toothbrushing and the participants were trained to assist where possible in the routine. In the Jarman 1983 study, training in five other self-care routines was included in the intervention.

The duration of the training for people with ID tended to be considerably longer than that for carers; the shortest period was 2 weeks (Lange 1985) and the longest 19 months (Snell 1989). However, this varied from participant to participant depending on their particular needs and some interventions included a maintenance phase, where the trainer prompted the participant to undertake the next step in the routine.

The description of the toothbrushing routine varied from "task completion" (Albino 1979; Jarman 1983), to broad stages (Lange 1985), to detailed step by step descriptions (Abramson 1972; Bouter 1979; Snell 1989). Disclosing tablets, to indicate plaque, were used as a teaching and monitoring aid in Lange (1985). Praise was used in all the interventions as a motivator; three studies also included physical rewards or tokens (Abramson 1972; Albino 1979; Jarman 1983). The training was led or delivered by a psychologist in four studies (Abramson 1972; Albino 1979; Bouter 1979; Jarman 1983), by a dentist in one study (Lange 1985) and by a teacher in another (Snell 1989).

Miscellaneous interventions

Three RCTs compared either brushing daily to brushing twice weekly or once weekly (Swallow 1969), dental recall intervals (one-monthly, three-monthly, six-monthly and usual care) (Shaw 1991) or using an individual's clinical photographs to discuss oral hygiene performance versus not using them (Bickley 1990). Two NRS compared using either a toothpaste with a plaque disclosing agent versus a conventional toothpaste (Teitelbaum 2009) or the use of an individualised oral care plan versus no individualised oral care plan (Altabet 2003).

Characteristics of outcomes:

Gingival health

Gingival inflammation

Twelve RCTs reported gingival inflammation as an outcome, 11 of which used a validated gingival index. The indices used were Löe & Silness (Carr 1997; Garcia-Carrillo 2016; Lange 1985; Phlypo 2016; Swallow 1969), Lobene (Mac Giolla Phadraig 2015), WHO Technical Report 621 Gingival Index (Shaw 1983), Saxer & Muhlemann Papillary Bleeding Index (Christen 2007; Kaschke 2005; Sauvetre 1995) and Ainamo & Bay (Bildt 2010). The 12th study used the subjective rating of "worse, same or better" (Kelner 1963).

Six NRS reported gingival inflammation as an outcome, two studies used a validated gingival index; Löe & Silness (Bratel 1991) and the WHO Technical Report 621 Gingival Index (Shaw 1991). The four studies using a subjective rating for gingival inflammation used the terms "obvious gingivitis, decrease in gingivitis or absence of clinically observable gingivitis" (Gertenrich 1967a; Gertenrich 1967b; Gertenrich 1967c; Gertenrich 1967d).

Modifications to the gingival indices were made in a number of studies, the most common modification was in relation to the number of teeth and surfaces examined. Thirteen studies used six standard teeth on the following surfaces (Table 14).

Table 14: Modifications to the Gingival Inflammation indices used

Surfaces assessed on six standard teeth for gingival inflammation	Studies
Four buccal and two lingual surfaces	Bildt 2010; Carr 1997; Mac Giolla Phadraig 2015; Phlypo 2016; Teitelbaum 2009
The Bentley 1995 modification; two randomly selected (by coin toss) quadrants (one in the upper jaw, one in the lower jaw, contralateral)	Garcia-Carrillo 2016
Four surfaces per tooth	Bratel 1991

Three surfaces per tooth; buccal, mesial and lingual	Sauvetre 1995
Two surfaces per tooth; on the buccal and lingual surfaces	Christen 2007; Kaschke 2005
Two surfaces per tooth; on the mesial and distal surfaces of each tooth	Lange 1985; Lange 2000
The interdental papillae and the intervening gingiva on the buccal surfaces of all standing teeth	Swallow 1969
Other modifications	Studies
No probe used and only teeth that were visible to the examiner	Mac Giolla Phadraig 2015
If standard teeth were missing, they were not substituted	Lange 2000

Only two studies reported carrying out a periodontal pocket index assessment (WHO 1978) and only one of these, an NRS, reported the findings (Shaw 1991).

Plaque

Fourteen RCTs reported plaque as an outcome using a validated index. The indices used were: Silness & Loe (Garcia-Carrillo 2016; Phlypo 2016; Sauvetre 1995; Shaw 1983; Swallow 1969), Green & Vermillion (Bildt 2010; Carr 1997; Lange 1985), Quigley & Hein (Christen 2007; Dôgan 2004; Kaschke 2005), Kobayashi & Ash (Albino 1979, the WHO Technical Report No 621 (Bickley 1990), Approximal Plaque Index (Christen 2007; Dôgan 2004; Kaschke 2005) and Shaw & Murray (Bickley 1990). One RCT used a subjective rating of oral hygiene describing the outcome using the terms "worse", "same" or "better" (Kelner 1963).

Six NRS reported plaque as an outcome using a validated index; the indices used were: Silness & Loe (Bratel 1991), Green & Vermillion (Glassman 2006; Teitelbaum 2009; Williams 1988), Ramfjord (Lange 2000) and the WHO

Technical Report No 621 (Shaw 1991). A further five NRS used a subjective rating describing the outcome using the following terms: "excellent", "good", "fair", "poor" or "very poor" (Altabet 2003); "good", "fair", "poor" or "very poor" (Gertenrich 1967a; Gertenrich 1967b; Gertenrich 1967c; Gertenrich 1967d).

Four studies used more than one measure of plaque. Modifications to the indices were made in a number of studies, most commonly in relation to the number of teeth and surfaces examined. Sixteen studies used six standard teeth on the following surfaces (Table 15).

Table 15: Modifications to the Plaque indices used

Surfaces assessed on six standard teeth for plaque levels	Studies
Four buccal and two lingual surfaces	Bickley 1990; Carr 1997; Glassman 2006; Mac Giolla Phadraig 2015; Phlypo 2016; Shaw 1983; Shaw 1991; Teitelbaum 2009; Williams 1988
The Bentley 1995 modification; two randomly selected (by coin toss) quadrants (one in the upper jaw, one in the lower jaw, contralateral)	Garcia-Carrillo 2016
Four surfaces per tooth	Bratel 1991
Three surfaces per tooth; buccal, mesial and lingual	Sauvetre 1995
Two surfaces per tooth; on the buccal and lingual surfaces	Albino 1979; Bildt 2010
Two surfaces per tooth; on the mesial and distal surfaces of each tooth	Lange 1985; Lange 2000
Other modifications	Studies
No probe used and only teeth that were visible to the examiner	Mac Giolla Phadraig 2015
If standard teeth were missing, they were not substituted	Lange 2000

Calculus

Four studies reported on calculus; three RCTs and one NRS. The indices used were: Green & Vermillion (Carr 1997; Garcia-Carrillo 2016) and the WHO Technical Report 621 (Bickley 1990 (assessed but not reported); Shaw 1991).

All three studies that reported their findings used the six standard teeth (Shaw 1991; Carr 1997; Garcia-Carrillo 2016). Two studies assessed four buccal and two lingual surfaces and one study assessed four surfaces per tooth (buccal, lingual, mesial and distal).

Oral assessment

Ferozali 2007 used a tool to assess the health of four elements; lips oral mucosa, gingivae and plaque, which was a modification of the Beck Total Oral Assessment Tool. This study also measured a total bacteria count and a potentially pathogenic bacteria count.

The following are the references for the clinical indices referred to in this section (Characteristics of outcomes): Ainamo & Bay 1975; Beck & Yasko 1993; Bentley & Disney 1995; Fleiss *et al* 1987; Greene & Vermillion 1960; Kobayashi & Ash 1964; Lange *et al* 1977; Lobene *et al* 1986; Löe 1967; Saxer & Mühlemann 1975; Shaw & Murray 1977; WHO 1978.

Knowledge, behaviour, attitude and self-efficacy:

Knowledge

Three RCTs assessed changes in oral health care knowledge of carers, all using multiple-choice questionnaires, two of which were specifically designed by the investigators for the intervention (Mac Giolla Phadraig 2015; Gonzalez 2013), and the third (Phlypo 2016) used the questionnaire designed by Mac Giolla Phadraig (2015). Two studies had 10 questions on oral health-related knowledge (Mac Giolla Phadraig 2015; Phlypo 2016) and one study had 20 basic oral hygiene questions (Gonzalez 2013).

The questionnaire utilized by Mac Giolla Phadraig (2015) was reported as having been tested for reliability, validity, responsiveness and reproducibility and found to be acceptable. In the Gonzalez study (2013), a different questionnaire was used pre and post intervention, with similar content but different wording; the study authors sought expert advice to assess the content.

Behaviour, attitude and self-efficacy

Two of the studies that assessed knowledge of carers also assessed changes in their behaviour, attitude and/or self-efficacy at the same time: the studies had four questions on behaviour and five each on attitude and self-efficacy (Mac Giolla Phadraig 2015; Phlypo 2016). One study had assessed changes in self-reported behaviour alone, using five question (Olmos 2016). Four other NRS assessed behavioural changes in both carers and people with ID by monitoring of their behaviour using trained observers aided by specifically designed checklists (Bouter 1979; Glassman 2006; Jarman 1983; Kissel 1983). Reliability of the recorded observations was assessed by the study authors and was found to be acceptable in all four of these studies.

The methods used to assess changes in behaviour included: use of different methods of reinforcement by carers (Jarman 1983), assessing staff use of training methods when providing oral hygiene support (verbal instruction, physical guidance and reward) and participants' levels of self-initiation and their need for verbal instruction and physical guidance (Kissel 1983), the effect of training on carers and levels of feedback and support to them (Abramson 1972), behavioural methods used by carers to assist people with ID (Bouter 1979), the type of toothbrush used, the type of toothpaste used, the frequency of fluoride application, the timing of toothbrushing and duration of toothbrushing (Olmos 2016), caregivers' presence during toothbrushing (Glassman 2006) and the number of training sessions required for the toothbrushing skill to be performed without prompting (Snell 1989).

Indirect measures of behaviour change included measuring wear and tear on toothbrushes and a diary kept by the parents/staff of the participants' brushing frequency (Bratel 1991).

Unintended effects:

Adverse effects

No studies reported any positive unintended effects. Twenty-four studies did not report any unintended negative effects. Negative unintended or adverse effects were investigated or reported in 10 studies.

The adverse effects reported included discomfort when brushing, difficulty controlling the force and position of the electric toothbrush and the electric toothbrush being out of order (Bratel 1991), participants being frightened when the electric toothbrush was first used (Gertenrich 1967a), participant feeling sick (felt by the researchers to be a delaying tactic by the participant) (Bouter 1979), participants displaying sensitivity to mouths being held open (Snell 1989) and the bulkiness of the three-headed toothbrush causing some problems for those with limited opening (Christen 2007). One study reported participants being dismissed from the study, but no clear reason was provided (Carr 1997). Other potential adverse effects investigated but not found were allergy to the materials being used (Ferozali 2007), stress and fatigue following completion of a questionnaire (Gonzalez 2013), visual inspection for adverse effects from the toothbrushes being used (Garcia-Carrillo 2016).

Other outcomes

Other subjective outcomes were reported narratively, in relation to possible benefits or harm, for example, interviews with those using an electric toothbrush to assess their experiences using it (Bratel 1991), feedback from carers on the toothbrushes being used (Gertenrich 1967a; Gertenrich 1967b; Gertenrich 1967c; Gertenrich 1967d), social validation of participants' abilities with regard to oral hygiene completed by their carers and carers perception of training (Glassman 2006), a staff acceptability questionnaire (Kissel 1983) and questions on participants' skill retention (Snell 1989).

COM-B system characteristics

Most studies did not define behavioural change theories in the interventions. We therefore assessed all of the studies using the COM-B System to identify the behavioural change techniques inherent in the interventions (Michie *et al* 2011). We considered how the interventions affected capability (physical strength, skill or stamina or psychological strength, knowledge, skill or stamina), opportunity (physical opportunity afforded by the environment or the social opportunity afforded by social factors such as cultural norms or social cues) and motivation (reflective brain process; evaluation and plans or automatic brain processes; desires, impulses, inhibitions etc).

The element identified most often was capability: 29 studies were identified as having the potential to change the capability of either participants (11) or carers (8) or both (10), either on a physical (11) or on a physical and psychological level (18). These interventions included a focus on training, modelling or enablement of the participant or carer in the skill of toothbrushing.

In relation to the element of opportunity, physical opportunity was the more common potential element identified (26 studies). These interventions included elements such as specific time being set aside for the toothbrushing routine, environmental restructuring such as providing different oral hygiene or other aids or changing the setting where the toothbrushing took place. The potential for social opportunity (for example cultural norms or social cues) to impact on behaviour was identified in seven studies, which were predominantly targeted at carers and involved monitoring of their behaviour.

Nineteen studies had the potential for reflective motivation and these tended to include some element of feedback in relation to behaviour or performance which was seen predominantly in the interventions targeting carers. No studies were identified as having the potential for the behaviour change to become automatic.

We determined that 16 of the 34 studies had all three elements of capability, opportunity and motivation inherent in the intervention. The behavioural change elements and intervention functions are detailed in the individual Characteristics of included studies tables (Appendix 5).

Stakeholder involvement

The involvement of stakeholders may impact on the success of an intervention and so it was felt important to this review to capture the nature of the stakeholder's involvement in the interventions. Most of the studies actively involved parents or carers of the participants in the intervention (29 studies). Six studies involved both formal and non-formal carers, 22 studies involved formal carers only and one study used non-formal carers only. Three other studies were unclear if carers were involved. These studies were set in a school or day centre (Bickley 1990; Dôgan 2004; Sauvetre 1995). Two studies used

a member of the research team to brush the participants teeth (Swallow 1969; Williams 1988). Other stakeholders included administration staff, management staff, nursing or medical staff, speech and language therapists, an occupational therapist, a physical therapist and teachers. Eight studies reported no involvement of stakeholders, other than those directly involved in the research.

Twenty-six studies had dental professionals on the research team, 21 studies involved dentists or dental students, 16 studies involved dental hygienists or therapists, four studies involved dental nurses or assistants and one study involved an oral health promotor. The dental professionals were mainly involved in the initial oral hygiene instruction and the clinical measurements throughout the intervention. Seven studies indicated that the dental professional provided ongoing support during the intervention (Albino 1979; Bickley 1990; Glassman 2006; Sauvetre 1995; Shaw 1991; Swallow 1969; Williams 1988); one other indicated that the support was offered but not if the offer was taken up (Mac Giolla Phadraig 2015).

Funding

Nine studies reported receiving funding from their institution or health service (Abramson 1972; Bratel 1991; Gonzalez 2013; Jarman 1983; Olmos 2016; Kissel 1983; Mac Giolla Phadraig 2015; Shaw 1991; Teitelbaum 2009). Sixteen studies did not report on funding. Eight studies reporting receiving some funding from dental companies; all funding was stated as relating to the supply of either the toothbrushes or toothpaste required for the study (Albino 1979; Bickley 1990; Dôgan 2004; Garcia-Carrillo 2016; Kelner 1963; Sauvetre 1995; Shaw 1991; Swallow 1969).

4.6.4 Excluded studies

Following full-text reading, we rejected 158 reports for the following reasons: not an eligible study design (75), not an oral hygiene intervention (39), not mechanical removal of plaque (16), a complex intervention in which the oral hygiene component could not be appraised separately (11) and not relating to a person with ID or their carer (17).

We excluded a further 20 reports after further consideration for the following reasons. We excluded two as only the title was available (Borglin 1969; Lopez 1994) and a further five as only the abstract was available (Andrews 1990; Badra 1973; Ojeda 2010; Ribeiro 2011; Thornton 1991) and multiple efforts to contact the authors and source the full text were unsuccessful. A further three reports were excluded as there were no data presented in the published reports or data were presented in a format that could not be used and attempts to contact the authors were unsuccessful (Brody 1975; Favell 1975; Meador 1979). The authors of three other reports with data in a format that could not be used were contacted but the data were no longer available (Gertenrich 1972; Lesmana 2014; Schmidt 1981). Two other reports were trial registers, one had been written up in a thesis but it was not available and all attempts to contact the author were unsuccessful (ISRCTN10044161) and the authors of the second trial reported that the trial had not proceeded as yet, due to difficulties in recruiting participants (NCT03234231). One report was excluded as it was unclear if it met the inclusion criteria in relation to study design; all attempts to contact the authors were unsuccessful (Horner 1975). Three studies did not meet the required study design (Kaschke 2008; Haran 2014; Zaksek 2014). One of the authors of another study, with a subgroup of participants that may have been eligible for inclusion, confirmed that the study did not meet the study design criteria (Bui 2003) (see Characteristics of excluded studies: Appendix 7).

Two studies are awaiting classification (Birani *et al* 2008; Akgun *et al* 2012). We contacted the authors as the theses for both are available, but, despite reminders, the information was not received before the deadline indicated for inclusion in this review.

4.6.5 Risk of bias in included studies

We assessed the RCTs and NRCTs for risk of bias using standardised risk of bias criteria (Higgins *et al* 2011a). We assessed the ITS and RM studies using the EPOC seven standard criteria (EPOC 2016) (Figure 4; Figure 5).

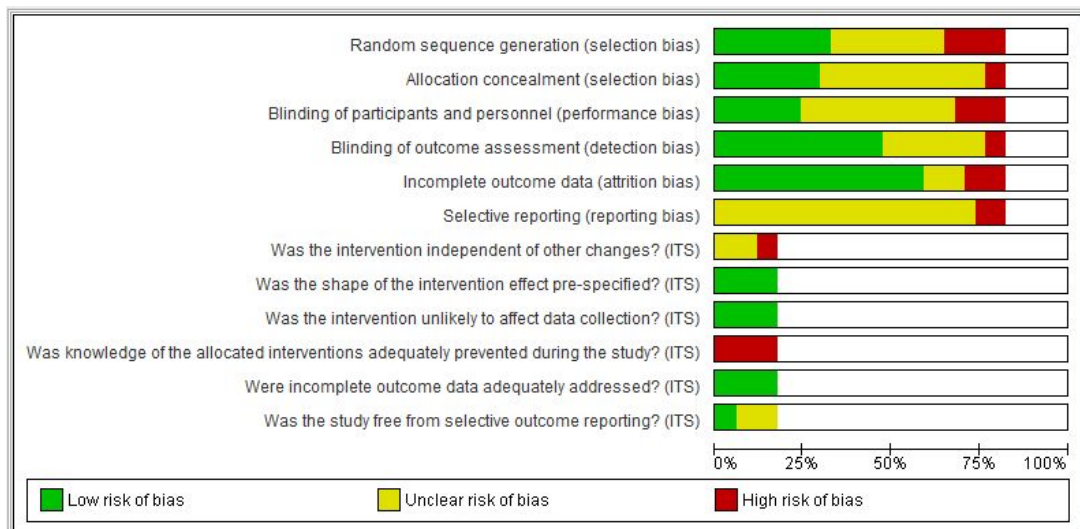


Figure 4: Cochrane Review Overall Risk of Bias

Of the 28 RCTs and NRCTs assessed, none were assessed as being at overall low risk of bias. Seventeen were assessed as being at overall high risk of bias (Albino 1979; Altabet 2003; Bickley 1990; Bratel 1991; Christen 2007; Dôgan 2004; Gertenrich 1967a; Gertenrich 1967b; Gertenrich 1967c; Gertenrich 1967d; Kaschke 2005; Kelner 1963; Lange 1985; Lange 2000; Mac Giolla Phadraig 2015; Phlypo 2016; Shaw 1983; Williams 1988); the remaining 10 were assessed overall as being at unclear risk of bias.

All of the ITS and RM studies were assessed overall as being at high risk of bias (Abramson 1972; Bouter 1979; Glassman 2006; Jarman 1983; Kissel 1983; Snell 1989).

Allocation (selection bias)

Of the 19 RCTs included in this review, eleven were regarded as being at low risk of bias in relation to random sequence generation (Albino 1979; Bickley 1990; Bildt 2010; Christen 2007; Ferozali 2007; Garcia-Carrillo 2016; Gonzalez 2013; Mac Giolla Phadraig 2015; Shaw 1983; Swallow 1969; Sauvetre 1995). These studies clearly indicated the method used for randomisation. A further ten RCTs stated that the participants were randomised but gave no details of how this randomisation process was carried out. These studies were rated as of 'unclear' risk of bias. No RCTs were assessed as being at high risk of bias. The allocation concealment rating for

the RCTs was nine studies at low risk of bias and ten studies were at 'unclear' risk of bias.

The nine NRCTs were assessed using the RCT risk of bias criteria (Higgins *et al* 2011a). Six of these studies were assessed as being at 'high' risk of bias for random sequence generation as the participants were selected non-randomly or the participants were selected for particular groups, the remaining three studies were assessed as being at 'unclear' risk of bias as no reference to randomisation was made. One NRCT was assessed as being at 'low' risk of bias (Lange 2000), two of the NRCT were assessed as being at 'high' risk of bias for allocation concealment as (Altabet 2003; Bratel 1991). The remaining six NRCTs were assessed as being at 'unclear' risk of bias for allocation concealment as no details were provided in the reports in relation to this element.

Blinding (performance bias and detection bias)

Four of the RCTs were assessed as being at 'high' risk of bias in relation to blinding of participants and/or personnel: all four were cross-over trials (Christen 2007; Dôgan 2004; Kaschke 2005; Shaw 1983). Five RCTs were assessed as being at low risk as the control group were asked to do something that was not usual care (Bildt 2010; Gonzalez 2013; Kelner 1963; Sauvetre 1995; Shaw 1991). The remaining ten RCTs were at 'unclear' risk of bias.

One NRCT was assessed as being at 'high' risk of bias in relation to blinding of participants and/or personnel (Williams 1988). Three NRCTs were assessed as being at low risk of bias as some attempt had been made to blind either the participants or personnel (Altabet 2003; Lange 2000; Teitelbaum 2009). The remaining six NRCTs were assessed as having 'unclear' risk of bias for blinding of participants and/or personnel (Bratel 1991; Gertenrich 1967a; Gertenrich 1967b; Gertenrich 1967c; Gertenrich 1967d).

In relation to blinding of outcome assessors in the RCTs, ten studies were assessed as being at low risk of bias as the assessors were blinded to the group allocation (Bickley 1990; Bildt 2010; Christen 2007; Ferozali 2007; Garcia-Carrillo 2016; Kaschke 2005; Kelner 1963; Sauvetre 1995; Shaw 1983; Swallow 1969); seven studies were at 'unclear' risk of bias as there were

insufficient details provided in the reports in relation to blinding (Carr 1997; Dôgan 2004; Gonzalez 2013; Mac Giolla Phadraig 2015; Olmos 2016; Phlypo 2016; Shaw 1991); two studies were assessed as being at 'high' risk of bias as the assessors were not blinded to the group allocation (Albino 1979; Lange 1985).

In the NRCTs, six studies were assessed as being at low risk of bias as assessors were blinded to the group allocation; the remaining three studies were at 'unclear' risk of bias as no mention of blinding of assessors was reported (Bratel 1991; Teitelbaum 2009; Williams 1988).

Incomplete outcome data (attrition bias)

Four RCTs were assessed as being at 'high' risk of bias in relation to incomplete outcome data as either there were 'high' attrition rates with no explanation as to the reasons for the drop outs (Mac Giolla Phadraig 2015; Phlypo 2016; Shaw 1983) or drop outs were due to lack of participation by the participants (Albino 1979). Three studies were assessed as having 'unclear' risk of bias as either there was insufficient detail in relation to the reason or number of drop outs (Dôgan 2004) or the reasons provided may have impacted on the outcomes: participants lost or broke the electric toothbrush (Carr 1997) or the participants were unable to following the training (Sauvetre 1995). The remaining twelve studies were assessed as being at low risk of bias for incomplete outcome data.

One NRCT was assessed at being at 'unclear' risk in relation to incomplete outcome data due to the lack of details provided (Gertenrich 1967a). The remaining eight NRCTs were at low risk of bias.

Selective reporting (reporting bias)

Three RCTs were assessed as being at 'high' risk of bias in relation to selective reporting: one because the plaque index was modified during the intervention as it proved too difficult to complete (Albino 1979); two because all the outcomes outlined in the methodology were not reported (Bickley 1990; Kelner 1963). Fourteen studies were assessed as having an 'unclear' risk of bias as there was no protocol for the study, but all listed outcomes were reported.

Another study referred to mid-term data, but they were not reported (Shaw 1983) and one study had a protocol, but it appeared to have been completed retrospectively (Garcia-Carrillo 2016). No studies were at low risk of bias in relation to selective reporting.

All nine NRCTs were assessed as being at 'unclear' risk of bias for selective reporting as there was no protocol but all the expected outcomes reported in the methodology sections were reported as planned.

Other potential sources of bias

No other potential sources of bias were identified.

Overall, there was a large element of 'unclear' risk of bias in many of the domains, reflecting the lack of detail provided in the published reports in relation to the methodology. This may be due to the large number of older studies included in this review. There were no studies at low risk of bias for the domain of selective reporting: many of the studies had no protocol and so were assessed as being at unclear risk of bias for this domain.

Risk of bias for Interrupted Time Series and Repeat Measure studies

Three interrupted time series studies (ITS) (Jarman 1983; Kissel 1983; Glassman 2006) and three repeat measure studies (RM) (Abramson 1972; Bouter 1979; Snell 1989) were assessed for risk of bias using the Effective Practice and Organisation of Care (EPOC) Group standards (EPOC 2016):

Was the intervention independent of other changes?

Of the six studies, two were assessed as being independent of other changes (low risk) (Bouter 1979; Glassman 2006); three were assessed as being unclear in relation to being independent of other changes as insufficient information was provided (Abramson 1972; Kissel 1983; Snell 1989); and one was assessed as not being independent (high risk), as the skill of toothbrushing was one of six other skills being taught at the same time (Jarman 1983).

Was the shape of the intervention effect prespecified?

All six ITS and RM studies had prespecified the shape of the intervention in relation to the methodology and expected outcomes (low risk).

Was the intervention unlikely to affect data collection?

The intervention in all six studies was assessed as being unlikely to affect data collection (low risk).

Was knowledge of the allocated interventions adequately prevented during the study?

It was not possible to prevent knowledge of the allocated intervention in the six studies as the assessors in all cases were actively involved in the delivery of the intervention (high risk).

Were incomplete outcome data adequately addressed?

Incomplete data were explained in two studies and assessed as not having an impact on the outcomes (Bouter 1979; Kissel 1983) and there were no incomplete data in the other four studies (low risk).

Was the study free from selective outcome reporting?

Five of the studies were assessed as being free from selective outcome reporting as all outcomes outlined in the methodology were reported (low risk). One study was assessed as being unclear as it was difficult to extract the data from the figures provided (Glassman 2006).

Was the study free from other risks of bias?

No other risks of bias were identified.

	Random sequence generation (selection bias)	Allocation concealment (selection bias)	Blinding of participants and personnel (performance bias)	Blinding of outcome assessment (detection bias)	Incomplete outcome data (attrition bias)	Selective reporting (reporting bias)	Was the intervention independent of other changes? (ITS)	Was the shape of the intervention effect pre-specified? (ITS)	Was the intervention unlikely to affect data collection? (ITS)	Was knowledge of the allocated interventions adequately prevented during the study? (ITS)	Were incomplete outcome data adequately addressed? (ITS)	Was the study free from selective outcome reporting? (ITS)
Abramson 1972							?					
Albino 1979	+	?	?	+	+	+						
Altabet 2003	+	+	+	+	+	?						
Bickley 1990	+	?	?	+	+	+						
Bildt 2010	+	+	+	+	+	?						
Bouter 1979							?	+	+	+	+	?
Bratel 1991	?	+	?	?	?	?						
Carr 1997	?	?	?	?	?	?						
Christen 2007	+	+	+	+	+	?						
Dögan 2004	?	?	+	?	?	?						
Ferozali 2007	+	+	?	+	+	?						
Garcia-Carrillo 2016	+	+	?	+	+	?						
Gertenrich 1967a	+	?	?	+	?	?						
Gertenrich 1967b	+	?	?	+	+	?						
Gertenrich 1967c	+	?	?	+	+	?						
Gertenrich 1967d	+	?	?	+	+	?						
Glassman 2006							?	+	+	+	+	?
Gonzalez 2013	+	+	+	?	+	?						
Jarman 1983							+	+	+	+	+	?
Kaschke 2005	?	?	+	+	+	?						
Kelner 1963	?	?	+	+	+	+						
Kissel 1983							+	+	+	+	+	?
Lange 1985	?	?	?	+	+	?						
Lange 2000	+	+	+	+	+	?						
Mac Giolla Phadraig 2015	+	+	?	?	+	?						
Olmos 2016	?	?	?	?	?	?						
Phypo 2016	?	?	?	?	+	?						
Sauvetre 1995	+	+	+	+	?	?						
Shaw 1983	+	+	+	+	+	?						
Shaw 1991	?	?	+	?	+	?						
Snell 1989							?	+	+	+	+	+
Swallow 1969	+	+	?	+	+	?						
Tettelbaum 2009	?	?	+	?	+	?						
Williams 1988	?	?	+	?	?	?						

Figure 5: Cochrane Review Individual Studies Risk of Bias

4.7 Effects of interventions

The intervention types (Comparisons), the outcomes assessed, as well as the time frames of the interventions, are summarized in the table below (Table 16). The detailed findings for each comparison are then presented in the succeeding pages. The certainty of the evidence for each outcome is categorised as high, moderate, low or very low. The grading of the risk of bias in the forest plots is colour coded: Low Risk – Green, Unclear Risk – Yellow, High Risk – Red.

Table 16: Summary table of Comparisons

No.	Comparison	Outcomes	Time frames
1	Special manual toothbrushes versus conventional manual toothbrushes for people with intellectual disabilities	Gingival inflammation	Short term, Medium term
		Plaque	Short term, Medium term
		Oral health assessment	Medium term
		Bacteria count	Medium term
		Behaviour	Medium term
2	Electric toothbrush versus manual toothbrush for people with intellectual disabilities	Gingival Inflammation	Short term, Medium term
		NRS Gingival inflammation	Medium term, long term
		Plaque	Short term, Medium term
		NRS Gingival inflammation	Medium term, long term
		Calculus	Medium term
3	Oral hygiene care training versus no oral hygiene care training or a placebo training session for carers of people with intellectual disabilities	Gingival inflammation	Medium term
		Plaque	Medium term
		NRS Plaque	Short term, Medium term

		Knowledge	Short term, Medium term
		Behaviour	Short term
		Behaviour, Attitude, self- efficacy	Medium term
4	Oral hygiene training versus no oral hygiene training of people with intellectual disabilities	Gingival inflammation	Short term
		Plaque	Short term
		Behaviour	Medium term, Long term
5	One, three- and six-monthly dental recall intervals versus no dental recall for people with intellectual disabilities	Gingival inflammation	Long term
		Gingival bleeding	Long term
		Plaque	Long term
		Calculus	Long term
6	The use of individual clinical photographs as a toothbrushing motivator versus no motivator for people with intellectual disabilities	Plaque	Medium term
7	Daily, twice weekly and once weekly frequencies of brushing versus usual care for people with intellectual disabilities	Gingival inflammation	Short term
8	The use of a toothpaste with a plaque disclosing agent versus a conventional toothpaste for people with intellectual disabilities	Gingival inflammation	Short term
		Plaque	Short term
9	An individualised oral care plan versus no oral care plan for people with intellectual disabilities	Plaque	Medium term

Comparison 1: Special manual toothbrushes versus conventional manual toothbrushes for people with ID

1.1 Gingival Inflammation (Short term / < six weeks)

Three RCTs, with 169 participants, compared the effect of using a special manual toothbrush with a conventional manual toothbrush on gingival inflammation over periods of two or three-weeks (Christen 2007; Kaschke 2005; Sauvetre 1995) (Table 17).

Two of the studies were three-armed cross-over trials, the third arm compared an electric toothbrush (reported separately). The special brush used in all three studies was the Superbrush. Only one of these studies presented the data in a format that could be entered on a forest plot (Sauvetre 1995).

This study showed no evidence of a difference between the two toothbrushes (MD -0.10, 95% CI -0.77 to 0.57) for people with ID in the short term (Figure 6). The certainty of the evidence is moderate. The two other studies had similar findings (Table 17).

1.2 Gingival Inflammation (Medium term / six weeks to twelve months)

One RCT compared the effect of using a special manual toothbrush (Superbrush) with a conventional manual toothbrush on gingival inflammation levels over a period of 4 months (Bildt 2010) (Table 18).

This study shows a small, statistically significant, effect size in favour of the special manual toothbrush for adults with intellectual disabilities in the medium term (four months) (MD: -12.40, 95% CI: -24.31 to -0.49), (Figure 7). The certainty of the evidence is low.

1.3 Plaque (short term / < 6 weeks)

Four RCTs with 189 participants, aged between four and 79 years, compared the effect of using a special manual toothbrush with a conventional manual

toothbrush on plaque levels over periods of 1, 2 and 3 weeks (Christen 2007; Dôgan 2004; Kaschke 2005; Sauvetre 1995). The special brush used in all four studies was the Superbrush. Three of the studies were three-armed cross-over trials; the third arm compared an electric toothbrush (reported separately) (Table 19).

One study that presented data that could be entered on a forest plot showed no evidence of a difference between the toothbrushes (MD 0.20 95% CI -0.45 to 0.85), (Figure 8). The certainty of the evidence is moderate. Of the three studies not included on the forest plot, two studies showed evidence in favour of the special manual toothbrush and the third study showed no difference between the toothbrushes

1.4 Plaque (medium term / 6 weeks to 12 months)

One RCT, with 18 participants, compared the effect of using a special manual toothbrush (Superbrush) with a conventional manual toothbrush on plaque levels, over a period of four months (Bildt 2010) (Table 20). The baseline scores were not equal, and the assessors were not calibrated introducing an element of indirectness in relation to certainty of the evidence. This study showed a moderate effect size in favour of a special manual toothbrush (Superbrush) for people with ID medium term (MD -0.44, 95% CI -0.93 to 0.05), (Figure 9). The certainty of the evidence is low.

One NRS found the advantage of using either a special (Collis Curve toothbrush) or a conventional manual toothbrush is uncertain (Table 21).

1.5 Oral health assessment and bacteria levels (medium term)

One RCT, with 22 participants, compared the effect of using a special manual toothbrush (single-use suction toothbrush) with a conventional manual toothbrush on listed elements of general oral health and on oral bacteria levels over a period of 90 days (Ferozali 2007) (Table 22). All the participants had their teeth brushed by a carer.

The effect sizes of these assessments were small or moderate (MD ranged from 0.12 to 0.45, 95% CI ranged from -0.67 to 0.91, to -1.35 to 0.45), (Figure

10). For people with intellectually disabilities, who have their teeth brushed by a carer, the effects of using either a single-use suction toothbrush or a conventional toothbrush, with a therapeutic chemical agent are equally effective over a period of 90 days. The certainty of the evidence is moderate.

1.6 Behaviour of people with ID and their carers (medium term)

One RCT compared the effect of using a special manual toothbrush (Superbrush) with a conventional manual toothbrush on the behaviour of the people with ID and their carers over a period of four months (Bildt 2010) (Table 23). Meta-analyses of the carer elements show no evidence of a difference between toothbrushes for the carers experience when handling the toothbrush, when brushing the teeth of people with ID or for the frequency of brushing by the carer, in the medium term (MD 0.50, 95% CI -0.35 to 1.35, MD 0.70, 95% CI -0.15, 1.55 and MD 0.20, 95% CI -2.13 to 2.53). (Figure 11).

The behaviour of people with ID (level of resistance to toothbrushing), showed a moderate but non-significant effect size, in favour of using the special manual toothbrush, medium term (MD -1.70, 95% CI -4.90 to 1.50), (Figure 12). The certainty of the evidence is low.

The effect estimates for all outcomes and subgroups included in forest plots for Comparison 1: Special manual toothbrushes versus conventional manual toothbrushes for people with intellectual disabilities, are presented in Table 24.

Table 17: C1 Gingival inflammation studies - short term

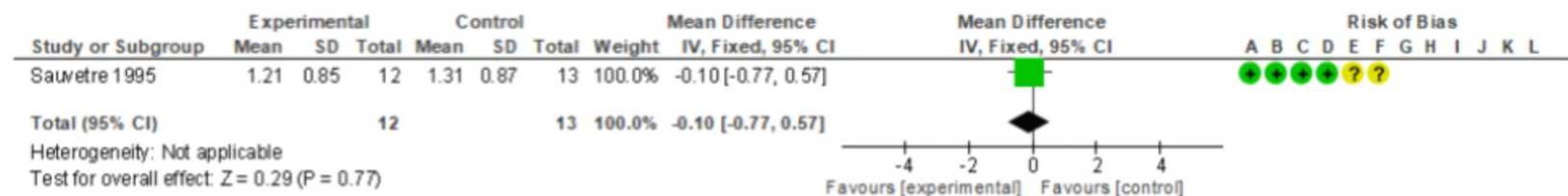
Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
2 weeks	Kaschke 2005	SuperBrush® versus Oral B Manual TB®	PBI (Scale 0-4)	Mouth Score 3.55 (0.40) Surface Score 7.1 (0.8)	Mouth Score 4.13 (0.35) Surface Score 8.25 (0.7)	54	Germany	Cross-over trial. Data are presented for the 2 arms combined. Surface Score Mean and SD obtained from author. Mouth Score calculated from this data. Median presented in published article.	The mean for the PBI for the Superbrush is below that of Manual TB. However, there was no strong evidence of difference between the two toothbrushes.
2 weeks	Christen 2007	SuperBrush® versus Oral B Manual TB®	PBI (Scale 0-4)	Pooled Median 12.2 Self Br 10.5 Assist Br 11.5 Other Br 14.5	Pooled Median 12.6 Self Br 10.0 Assist Br 12.5 Other Br 15.5	72	Germany	Cross-over trial. Data are presented for the 2 arms combined. Pooled median data calculated from reported data.	"Patients who brushed their teeth did not achieve significant changes in PBI levels with any of the three toothbrushes (Wilcoxon test: $p > 0.05$).\" Pg. 63.
21 days	Sauvetre 1995	Superbrush® (triple headed) versus standard manual brush (Oral B®).	S&MG I (Scale 0-3)	1.21 (0.85)	1.31 (0.87)	25	Belgium	Participant numbers not clearly reported, assumed to be 25.	"No significant difference was found when the results of bleeding indices were compared between the two toothbrushes\" Pg. 117

Footnotes

Kaschke 2005; Three-armed cross-over trial; also compared the Teledyne Waterpik Sonic Speed Electric TB, reported separately. Data for buccal and lingual surfaces, maxilla and mandible and self-brushers, assisted brushers and other brushers also reported. Christen 2007; Three-armed cross-over trial; also compared the Dentacare-Sonodont Electric TB, reported separately. Median and percentile data only reported. Data presented for self-brushers, assisted brushers and other brushers, pooled data have been calculated from these data. Data for buccal and lingual surfaces, maxilla and mandible also reported.

Indices: PBI; Papillary Bleeding Index. S&MG I - Saxer and Muhlemann Bleeding index 1975

1.1 Gingival Inflammation short term (< 6 weeks)



Risk of bias legend

- (A) Random sequence generation (selection bias)
- (B) Allocation concealment (selection bias)
- (C) Blinding of participants and personnel (performance bias)
- (D) Blinding of outcome assessment (detection bias)
- (E) Incomplete outcome data (attrition bias)
- (F) Selective reporting (reporting bias)
- (G) Was the intervention independent of other changes? (ITS)
- (H) Was the shape of the intervention effect pre-specified? (ITS)
- (I) Was the intervention unlikely to affect data collection? (ITS)
- (J) Was knowledge of the allocated interventions adequately prevented during the study? (ITS)
- (K) Were incomplete outcome data adequately addressed? (ITS)
- (L) Was the study free from selective outcome reporting? (ITS)

Figure 6: C1 Meta-analysis Gingival inflammation - short term

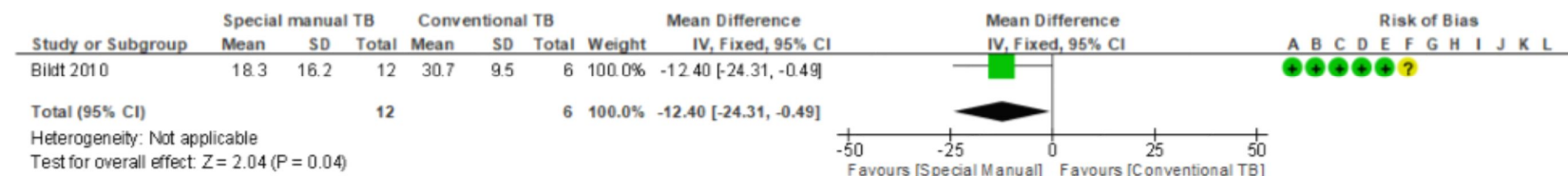
Table 18: C1 Gingival inflammation studies - medium term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
4 months	Bildt 2010	SuperBrush® versus Oral B Manual TB®	GBI (Scale 0-1)	18.3 (16.2)	30.7 (9.5)	18	Germany	No calibration of assessors, patients own dentist assessed clinical outcomes. The baseline scores were considerably lower for the intervention group (24.7 versus 32.7).	The author reports no strong evidence was found for a difference between the two toothbrushes for gingival inflammation.

Footnotes:

Indices: GBI – Gingival Bleeding Index Ainamo & Bay (0-1), 6 standard teeth, % of positive sites examined.

1.2 Gingival inflammation medium term (6 weeks to 12 months)



Risk of bias legend

- (A) Random sequence generation (selection bias)
- (B) Allocation concealment (selection bias)
- (C) Blinding of participants and personnel (performance bias)
- (D) Blinding of outcome assessment (detection bias)
- (E) Incomplete outcome data (attrition bias)
- (F) Selective reporting (reporting bias)
- (G) Was the intervention independent of other changes? (ITS)
- (H) Was the shape of the intervention effect pre-specified? (ITS)
- (I) Was the intervention unlikely to affect data collection? (ITS)
- (J) Was knowledge of the allocated interventions adequately prevented during the study? (ITS)
- (K) Were incomplete outcome data adequately addressed? (ITS)
- (L) Was the study free from selective outcome reporting? (ITS)

Figure 7: C1 Meta-analysis Gingival inflammation - medium term

Table 19: C1 Plaque studies - short term

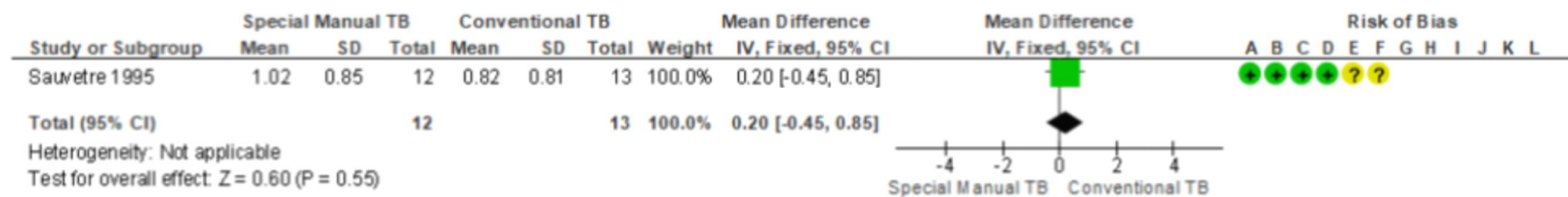
Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
1 week	Dôgan 2004	Superbrush® versus Oral B Cross Action 35/40 manual TB®	MQHPI (Scale 0-5)	1.77 (0.62)	2.15 (0.58)	15	Turkey	Cross-over trial. Data are presented for the final time point only.	There was strong evidence of a difference in PI between the SuperBrush and Oral B Cross Action brush (p < 0.01) favouring the SuperBrush
2 weeks	Christen 2007	SuperBrush® versus Oral B Manual TB®	MQHPI (Scale 0-5)	Pooled Median 27.7 Self Br 26.5 Assist Br 26.5 Other Br 30.0	Pooled Median 28.3 Self Br 26.0 Assist Br 28.0 Other Br 31.0	72	Germany	Cross-over trial. Data are presented for the 2 arms combined. Median and percentile only reported. Pooled median data calculated from reported data	There was no strong evidence of a difference in the QH median plaque score across all patients between the three different toothbrushes in the final examination (Friedman test: p > 0.05, Tab. 13). Pg. 55.
2 weeks	Kaschke 2005	SuperBrush® versus Oral B Manual TB®	MQHPI (Scale 0-5)	Mouth Score 2.98 (0.08) Surface Score 35.8(.92)	Mouth Score 3.24 (0.10) Surface Score 38.9 (1.2)	54	Germany	Cross-over trial. Data are presented for the 2 arms combined. Surface Score Mean and SD obtained from author. Mouth score calculated from this data. Median presented in published article. Data for sub-groups also presented.	The plaque score for the Superbrush lies below that of the conventional manual tooth brush. The strength of the evidence was strong.
21 days	Sauvetre 1995	Superbrush® (triple headed) versus standard manual brush (Oral B®).	S&LPI (Scale 0-3)	1.02 (0.85)	0.82 (0.81)	25	Belgium	Participant numbers not clearly reported: assumed to be 25.	"No significant difference was found between the two toothbrushes in the means of day 0, 7 or 21" Pg. 117

Footnotes

Dôgan 2004 Three-armed cross-over trial; also compared the Braun Plaque Control 3D Electric TB®, reported separately. Data for age groups, surfaces and approximal plaque also reported. Christen 2007; Three-armed cross-over trial; also compared the Dentacare-Sonodont Electric TB, reported separately. Median and percentile data only reported. Data presented for self-brushers, assisted brushers and other brushers, pooled data have been calculated from these data. Data for buccal and lingual surfaces, maxilla and mandible also reported. Kaschke 2005; Three-armed cross-over trial; also compared the Teledyne Waterpik Sonic Speed Electric TB, reported separately. Data for buccal and lingual surfaces, maxilla and mandible and self-brushers, assisted brushers and other brushers also reported. Some strong evidence of differences was reported in favour of the SuperBrush in some of these sub groups.

Indices: MGHPPI: Modified Quigley & Hein Plaque Index; Scale of 0 - 5. S&LPI - Silness and Loe Plaque Index; Scale 0-3.

1.3 Plaque short term (< 6 weeks)



Risk of bias legend

- (A) Random sequence generation (selection bias)
- (B) Allocation concealment (selection bias)
- (C) Blinding of participants and personnel (performance bias)
- (D) Blinding of outcome assessment (detection bias)
- (E) Incomplete outcome data (attrition bias)
- (F) Selective reporting (reporting bias)
- (G) Was the intervention independent of other changes? (ITS)
- (H) Was the shape of the intervention effect pre-specified? (ITS)
- (I) Was the intervention unlikely to affect data collection? (ITS)
- (J) Was knowledge of the allocated interventions adequately prevented during the study? (ITS)
- (K) Were incomplete outcome data adequately addressed? (ITS)
- (L) Was the study free from selective outcome reporting? (ITS)

Figure 8: C1 Meta-analysis Plaque - short term

Table 20: C1 Plaque studies - medium term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
4 months	Bildt 2010	SuperBrush® versus Oral B Manual TB®	SG&VD (Scale 0-3)	Mouth Score 0.78 (0.7) 4.7 (4.2)	Mouth Score 1.22 (0.36) 7.3 (2.2)	18	Germany	Whole mouth score calculated from tooth scores. No calibration of assessors, patients own dentist assessed clinical outcomes. The baseline score was lower for the intervention group (8.3 versus 9.2)	Author reports no strong evidence found for a difference between the two toothbrushes for plaque (F (2,15) = 1.20), p = .33) Thesis Pg. 5.

Footnotes

Indices: SG&VDI: Simplified Green & Vermillion Debris Index; Scale 0 - 18, total of 0-3 for 6 standard teeth

Table 21: C1 Plaque NRS - medium term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
3 months	Williams 1988	Curved- bristle manual toothbrush (Collis Curve) versus conventional manual toothbrush	SG&VOHI (Scale 0-3)	1.25	1.03	24	USA	No SD reported. Contact with author, data no longer available. Brushing was carried out by student dental assistants.	No strong evidence was found for a difference between the two toothbrushes for the oral hygiene index (t= -.31, p <.76).

Footnotes

Indices: SG&VOHI - Simplified Green & Vermillion Oral Hygiene Index - plaque only.

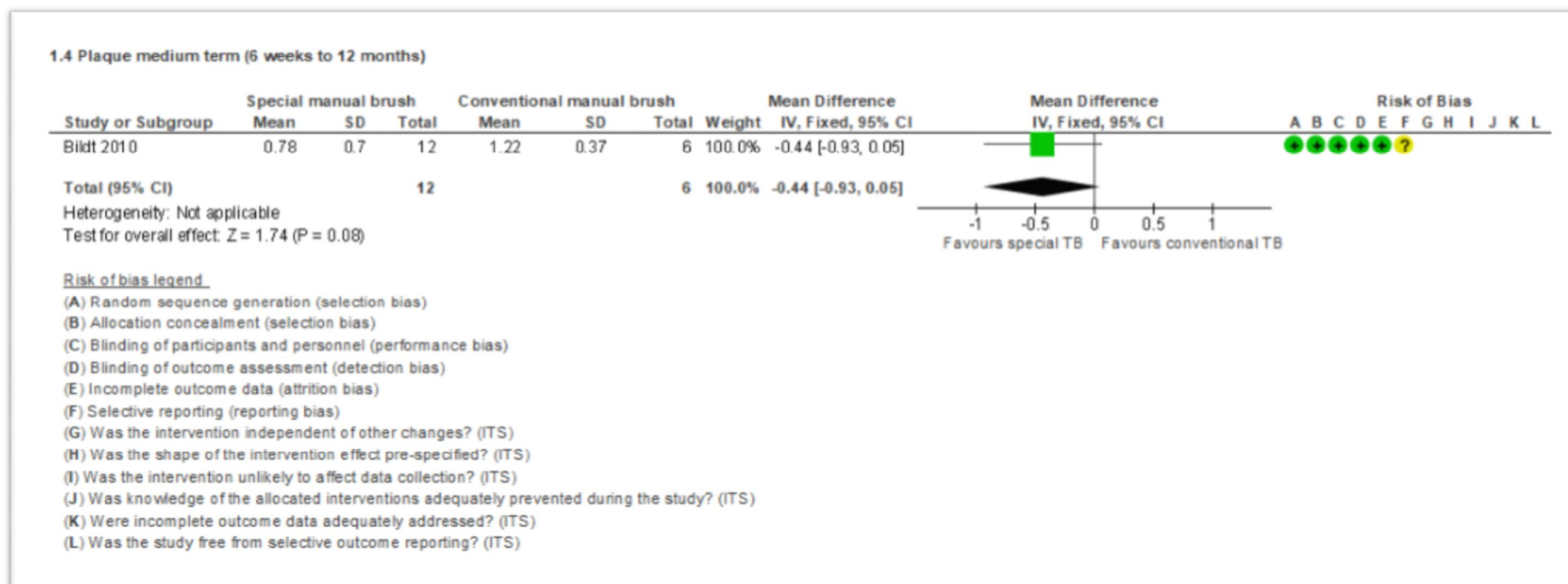


Figure 9: C1 Meta-analysis Plaque - medium term

Table 22: C1 Oral health assessment studies

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
90 days	Ferozali 2007	Oral cleansing with single use suction brush versus a standard manual toothbrush.	TOAS (Scale 0-12)	1.54 (1.0)	1.99 (1.12)	22	USA	Both groups brushed with an additional chemical agent. A third group not reported here used a standard toothbrush and toothpaste.	"There was a significant main effect over time of assessment, $F(2, 26) = 5.89$ ($p < 0.01$)" - all groups showed improvement. Participants receiving oral hygiene with suctioning "had the greatest improvements" in oral assessment scores and greatest sustainability over time. Pg. 173
90 days	Ferozali 2007	Oral cleansing with single use suction brush versus a standard manual toothbrush.	TBC	4.39 (0.63)	4.07 (0.80)	22	USA	Both groups brushed with an additional chemical agent. A third group not reported here used a standard toothbrush and toothpaste.	No strong evidence of a difference between the two groups, but there was a large drop in TBC in both groups, the suction toothbrush group reduced the TBC by almost 90%, the standard toothbrush group reduced the TBC by 81%.
90 days	Ferozali 2007	Oral cleansing with single use suction brush versus a standard manual toothbrush.	PPB (log base 10)	0.41 (0.97)	0.29 (0.92)	22	USA	Both groups brushed with an additional chemical agent. A third group not reported here used a standard toothbrush and toothpaste.	No significant changes in PPB between groups, all changes were reductions for pathogenic bacteria. Pg. 173

Footnotes

Indices: TOAS - Total Oral Assessment Score. TBC - Total Bacteria Count. PPB - Potentially pathogenic bacteria

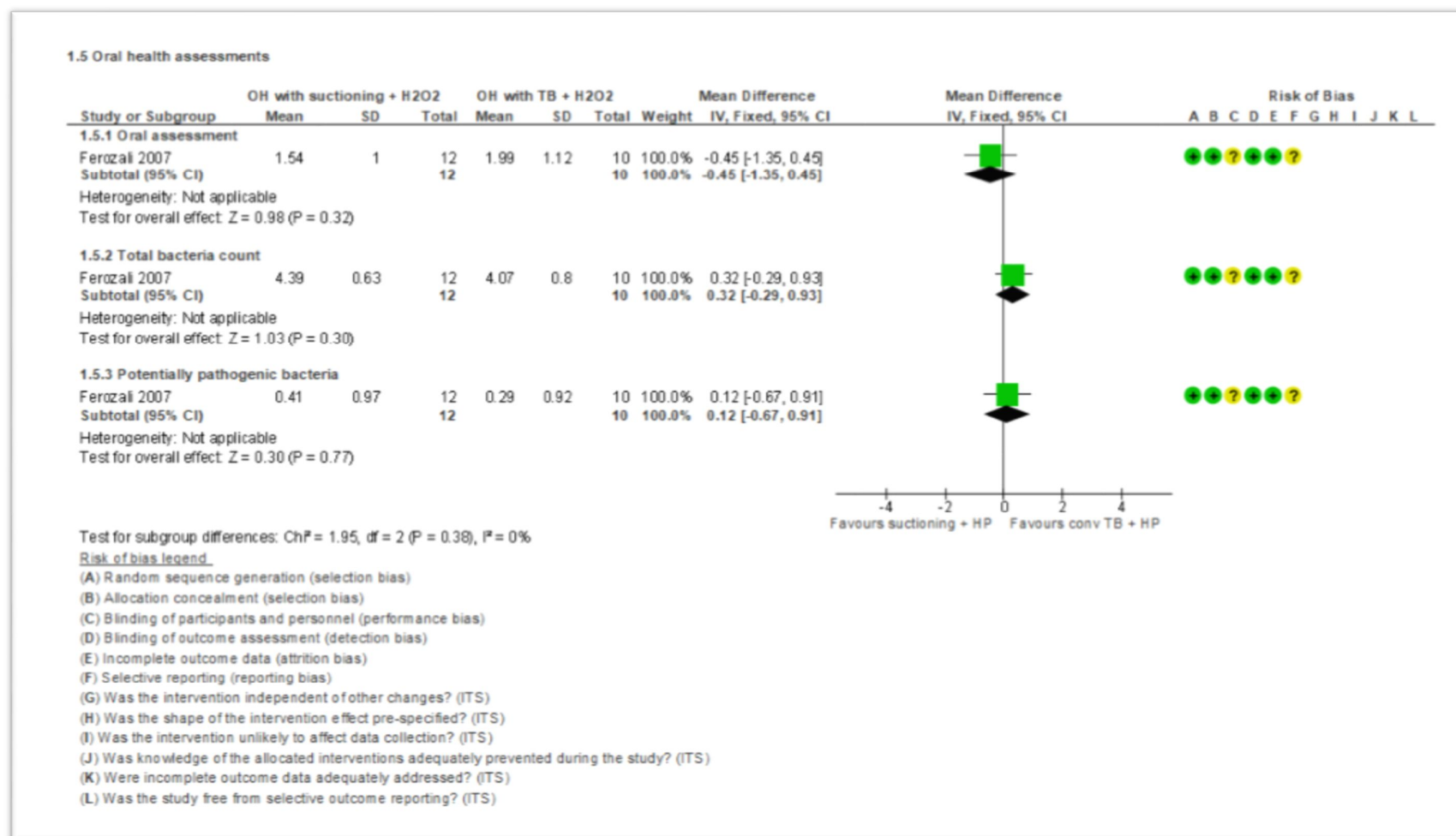
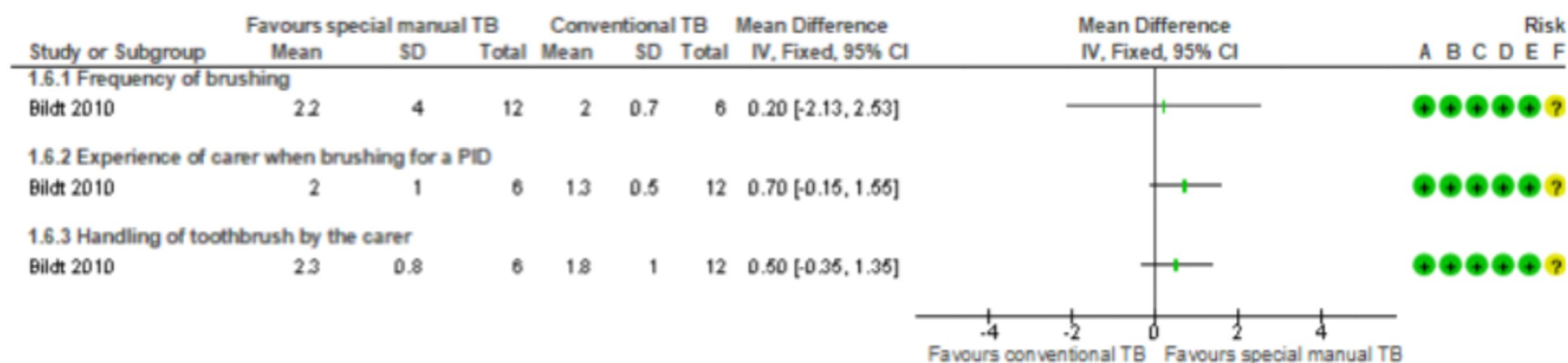


Figure 10: C1 Meta-analysis Oral health assessments

Table 23: C1 Behaviour studies - medium term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
4 months	Bildt 2010	SuperBrush® versus Oral B Manual TB®	Frequency of brushing by carer	2.0 (0.7)	2.2 (0.4)	18	Germany	Questionnaire was not validated	No strong evidence of a difference between toothbrushes
4 months	Bildt 2010	SuperBrush® versus Oral B Manual TB®	Level of resistance by PID	4.3 (3.2)	6.0 (3.4)	18	Germany	Questionnaire was not validated	No strong evidence of a difference between toothbrushes
4 months	Bildt 2010	SuperBrush® versus Oral B Manual TB®	Experience of carer when brushing	2.0 (1.0)	1.3 (0.5)	18	Germany	Questionnaire was not validated	No strong evidence of a difference between toothbrushes "although the experiences appeared to be more positive" for the carers using the special manual TB. Pg. 16
4 months	Bildt 2010	SuperBrush® versus Oral B Manual TB®	Handling of toothbrush by carer	2.3 (0.8)	1.8 (1.0)	18	Germany	Questionnaire was not validated	No strong evidence of a difference between toothbrushes

1.6 Behaviour, attitude and self-efficacy

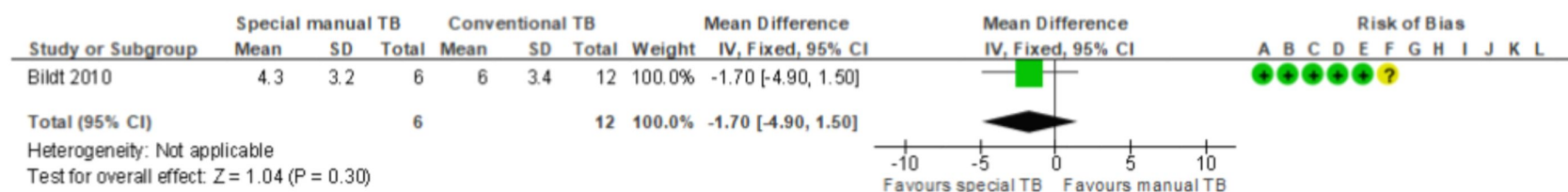


Risk of bias legend

- (A) Random sequence generation (selection bias)
- (B) Allocation concealment (selection bias)
- (C) Blinding of participants and personnel (performance bias)
- (D) Blinding of outcome assessment (detection bias)
- (E) Incomplete outcome data (attrition bias)
- (F) Selective reporting (reporting bias)
- (G) Was the intervention independent of other changes? (ITS)
- (H) Was the shape of the intervention effect pre-specified? (ITS)
- (I) Was the intervention unlikely to affect data collection? (ITS)
- (J) Was knowledge of the allocated interventions adequately prevented during the study? (ITS)
- (K) Were incomplete outcome data adequately addressed? (ITS)
- (L) Was the study free from selective outcome reporting? (ITS)

Figure 11: C1 Meta-analysis Behaviour – carer elements

1.7 Degree of resistance of PID



Risk of bias legend

- (A) Random sequence generation (selection bias)
- (B) Allocation concealment (selection bias)
- (C) Blinding of participants and personnel (performance bias)
- (D) Blinding of outcome assessment (detection bias)
- (E) Incomplete outcome data (attrition bias)
- (F) Selective reporting (reporting bias)
- (G) Was the intervention independent of other changes? (ITS)
- (H) Was the shape of the intervention effect pre-specified? (ITS)
- (I) Was the intervention unlikely to affect data collection? (ITS)
- (J) Was knowledge of the allocated interventions adequately prevented during the study? (ITS)
- (K) Were incomplete outcome data adequately addressed? (ITS)
- (L) Was the study free from selective outcome reporting? (ITS)

Figure 12: C1 Meta-analysis Degree of Resistance of people with ID

Table 24: Comparison 1 – All outcomes/ subgroups effect estimates

1 Special manual toothbrush versus conventional manual toothbrush (used by carers) for people with intellectual disabilities

Outcome or Subgroup	Studies	Participants	Statistical Method	Effect Estimate
1.1 Gingival inflammation short term (< 6 weeks)	1	25	Mean Difference (IV, Fixed, 95% CI)	-0.10 [-0.77, 0.57]
1.2 Gingival inflammation medium term (6 weeks to 12 months)	1	18	Mean Difference (IV, Fixed, 95% CI)	-12.40 [-24.31, -0.49]
1.3 Plaque short term (< 6 weeks)	1	25	Mean Difference (IV, Fixed, 95% CI)	0.20 [-0.45, 0.85]
1.4 Plaque medium term (6 weeks to 12 months)	1	18	Mean Difference (IV, Fixed, 95% CI)	-0.44 [-0.93, 0.05]
1.5 Oral health assessments	1		Mean Difference (IV, Fixed, 95% CI)	Subtotals only
1.5.1 Oral assessment	1	22	Mean Difference (IV, Fixed, 95% CI)	-0.45 [-1.35, 0.45]
1.5.2 Total bacteria count	1	22	Mean Difference (IV, Fixed, 95% CI)	0.32 [-0.29, 0.93]
1.5.3 Potentially pathogenic bacteria	1	22	Mean Difference (IV, Fixed, 95% CI)	0.12 [-0.67, 0.91]
1.6 Behaviour, attitude and self-efficacy	1		Mean Difference (IV, Fixed, 95% CI)	Subtotals only
1.6.1 Frequency of brushing	1	18	Mean Difference (IV, Fixed, 95% CI)	0.20 [-2.13, 2.53]
1.6.2 Experience of carer when brushing for a PID	1	18	Mean Difference (IV, Fixed, 95% CI)	0.70 [-0.15, 1.55]
1.6.3 Handling of toothbrush by the carer	1	18	Mean Difference (IV, Fixed, 95% CI)	0.50 [-0.35, 1.35]
1.7 Degree of resistance of PID	1	18	Mean Difference (IV, Fixed, 95% CI)	-1.70 [-4.90, 1.50]

Comparison 2: Electric toothbrushes versus manual toothbrushes for people with ID

2.1 Gingival inflammation (short term)

Four RCTs, with 270 participants, compared the effect of using an electric toothbrush with using a manual toothbrush on gingival inflammation, over periods of 2, 3 or 4 weeks (Christen 2007; Kaschke 2005; Swallow 1969; Shaw 1983), (Table 25). Three of the studies were cross-over trials, two included a third arm, which also compared a special manual toothbrush.

No strong evidence of a difference was found between the electric and manual toothbrushes for levels of gingival inflammation short term in any of the studies. None of the studies presented data in a format that could be included in a meta-analysis. The certainty of the evidence is low.

2.2 Gingival inflammation (medium term)

Three RCTs, two of which were cluster RCTs, with 220 participants, compared the effect of using an electric toothbrush with a manual toothbrush on gingival inflammation levels over periods of 3, 4, 6 and 12 months (Carr 1997; Garcia-Carrillo 2016; Kelner 1963 (Table 26).

Two cluster RCT studies, with 120 participants (Clusters = 10), and with low heterogeneity ($\text{Chi}^2 = 0.35$, $\text{df} = 1$ ($P = 0.56$); $I^2 = 0\%$) were included in a meta-analysis and showed no evidence of a difference for gingival inflammation levels between toothbrushes at six months (MD: 0.02, 95% CI: -0.06 to 0.09), (Figure 13). The certainty of the evidence in the meta-analysis is moderate.

2.3 NRS Gingival inflammation (medium and long term)

Five NRS, with 212 participants, compared the effect of using an electric toothbrush with using a manual toothbrush on gingival inflammation levels over a period of eight weeks to 16 months (Bratel 1991; Gertenrich 1967a; Gertenrich 1967b; Gertenrich 1967c; Gertenrich 1967d) (Tables 27 & 28).

At the 20 weeks to 16-month time-point none of the studies found strong evidence of a difference in levels of gingival inflammation between the use of the electric or manual toothbrushes. One NRS showed no evidence of a difference for gingival inflammation at 16 months (MD 0.00, 95% CI -0.15 to 0.15), (Figure 14). The certainty of the evidence is very low. The influence of other issues such as the level of ID, who was doing the brushing and the levels of assistance provided confounded the findings.

2.4 Plaque (short term/ < 6 weeks)

Four RCTs, with 270 participants, compared the effect of using an electric toothbrush with using a manual toothbrush on plaque levels, for people with ID, over periods of 1, 2 or 4 weeks (Christen 2007; Dôgan 2004; Kaschke 2005; Shaw 1983), (Table 29). All the studies were cross-over trials, three included a third arm, which also compared a special manual toothbrush (reported earlier).

None of the studies presented data in a format that could be included in a meta-analysis. Two studies showed evidence in favour of the electric toothbrush short term, the remaining two studies showed no evidence of a difference between the two toothbrushes. The benefit of using an electric toothbrush versus a conventional manual toothbrush short term is unclear, the certainty of the evidence is very low.

2.5 Plaque (medium term/ 6 weeks to 12 months)

Three RCTs, two of which were cluster RCT, with 220 participants, compared the effect of using an electric toothbrush with a manual toothbrush, for people with ID, on plaque levels over periods of 3, 4, 6 and 12 months (Carr 1997; Garcia-Carrillo 2016; Kelner 1963), (Table 30).

Two cluster RCT studies with 111 participants, with low heterogeneity ($\text{Chi}^2 = 0.13$, $\text{df} = 1$ ($P = 0.72$); $I^2 = 0\%$) were included in a meta-analysis and showed a small benefit in favour of the manual toothbrush at six months (SMD: 0.30, 95% CI: -0.08 to 0.67), (Figure 15). The certainty of the evidence was determined to be moderate.

2.6 NRS Plaque (medium and long term)

Five NRS, with 193 participants, compared the effect of using an electric toothbrush with a manual toothbrush, for people with ID, on plaque levels, over a period of 8 weeks to 16 months (Bratel 1991; Gertenrich 1967a; Gertenrich 1967b; Gertenrich 1967c; Gertenrich 1967d), (Table 31 & Table 32).

The data provided at the longest time points by the five studies (ranging between 8 and 34 weeks), with moderate heterogeneity ($\text{Tau}^2 = 0.10$; $\text{Chi}^2 = 6.97$, $\text{df} = 4$ ($P = 0.14$); $I^2 = 43\%$) were included in a meta-analysis (SMD: 0.04, 95% CI: -0.38 to 0.46), (Figure 16) and showed little or no evidence of a difference between the electric and manual toothbrushes medium term. The certainty of the evidence is very low.

2.7 Calculus (medium term)

Three RCTs, two of which were cluster RCTs, with 220 participants, compared the effects of using an electric toothbrush with a manual toothbrush, for people with ID, on calculus levels over periods of 4, 6 and 12 months (Carr 1997; Garcia-Carrillo 2016; Kelner 1963), (Table 33).

Two cluster RCTs, with 120 participants ($N = \text{clusters } 10$) and low to moderate heterogeneity ($\text{Chi}^2 = 1.44$, $\text{df} = 1$ ($P = 0.23$); $I^2 = 30\%$) were included in a meta-analysis and showed no evidence of a difference for calculus levels between toothbrushes at six months (SMD -0.04, 95% CI -0.40 to 0.32), (Figure 17). The certainty of the evidence was determined to be moderate.

The effect estimates for all outcomes and subgroups included in meta-analyses or Forest Plots for Comparison 2: Electric toothbrushes versus manual toothbrushes for people with intellectual disabilities are presented in Table 34.

Table 25: C2 Gingival inflammation studies - short term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
2 weeks	Kaschke 2005	Teledyne Waterpik Sonic Speed Electric TB® versus Oral B Manual TB®	PBI (Scale 0-4)	Whole Mouth 3.9 (0.35) Surface Score 7.8 (0.7)	Whole Mouth 4.3 (0.35) Surface Score 8.25 (0.7)	54	Germany	Cross-over trial. Data are presented for the 2 arms combined. Surface score Mean and SD obtained from author. Data calculated from obtained data. Median data presented in published article.	Although there was a great reduction in inflammation for the intervention "No significance differences were found" for PBI between the two groups (Pg. 68).
2 weeks	Christen 2007	Dentacare-Sonodont Electric TB® versus Oral B Manual TB®	PBI (Scale 0-4)	Pooled Median 12.3 Self Br 9.5 Assist Br 13.5 Other Br 14.0	Pooled Median 12.6 Self Br 10.0 Assist Br 12.5 Other Br 15.5	72	Germany	Cross-over trial. Data are presented for the 2 arms combined. Pooled median data calculated from reported data.	There was no strong evidence of a difference in the PBI median at the final examination and at the initial examination (Friedman test: $p > 0.05$, Tab. 17). Pg. 62
21 days	Swallow 1969	Daily brushing; Ronson Electric TB® versus Colgate-Palmolive manual TB®.	B&LL&SGI (Scale 0 - 3)	Pooled 1.24 Male; Buccal 2.57 Labial 2.08 Female; Buccal 0.14 Labial 0.15	Pooled 2.43 Male Buccal 2.07 Labial 2.34 Female; Buccal 2.63 Labial 2.65	20	UK	Pooled mean calculated from reported data, no SD provided.	Both brushes seemed equally effective. "None of the other factors examined (except frequency) produced an F values approaching the 5% level of significance" Pg. 377.
4 weeks	Shaw 1983	Braun Electric TB® versus Oral B 30 manual TB®.	WHOGI (Scale 0 - 3)	1.7 (1.9)	1.6 (1.7)	106	UK	Cross-over Trial. Data are presented for the two arms combined.	No strong evidence of a difference in oral hygiene performance when comparing the use of electric and manual toothbrushes. "There were no differences between the two groups. ($t=0.0702$, d.f. =/-52, $p > 0.05$)." Pg. 5

Christen 2007; Three-armed cross-over trial; also compared the Superbrush®, reported separately. Median and percentile data only reported. Data presented for self-brushers, assisted brushers and other brushers, pooled data have been calculated from these data. Data for buccal and lingual surfaces, maxilla and mandible, also reported. Kaschke 2005; Three-armed cross-over trial; also compared the Superbrush®, reported separately. Data for buccal and lingual surfaces, maxilla and mandible and self-brushers, assisted brushers and other brushers also reported. Swallow 1969; Means were presented separately for labial and buccal surfaces for males and females with no SD, the pooled means have been calculated from these data. Female data at 21 days appears skewed. Study also compared brushing one a week and twice a week, reported separately.

Indices: PBI; Papillary Bleeding Index; Scale 0 - 4. B&LL&SGI; Buccal and Labial surfaces Loe & Silness; Scale 0 - 3. WHOGI: WHO technical Report No 621 Gingival Index; Scale 0 - 3

Table 26: C2 Gingival inflammation studies - medium term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
3 months	Carr 1997	Interplak Electric TB® versus an Oral B 40 manual TB®.	L&SGI (Scale 0 - 3)	Pooled 1.80 (0.32) Self Br 1.81 (0.31) Assisted Br 1.78 (0.33)	Pooled 1.84 (0.25) Self Br 1.81 (0.28) Assisted Br 1.90 (0.21)	56	USA	Pooled mean and SD calculated from the reported data. Cluster RCT	There was no strong evidence of a difference in gingival indices between toothbrushes at 3 months.
3 months	Garcia-Carillo 2016	Sonicare EasyClean, Philips Electric TB® versus Vitis Access, Dentaids manual TB®	BDL&SGI (Scale 0-3)	1.31(0.32)	1.24 (0.32)	64	Spain	The description of ID in this study appears to include some people with an IQ above 70.	There was no strong evidence of a difference in gingival indices between toothbrushes at 3 months.
4 months	Kelner 1963	Automated electric TB (Broxodont®) versus Conventional TB	SGR	% Improvement 53%	% Improvement 31%	100	USA	% Improvement calculated from the reported data.	"The automatic electric toothbrush appeared significantly superior to the conventional toothbrush in improving gingival health" Pg. 108
6 months	Carr 1977	Interplak Electric TB® versus an Oral B 40 manual TB®.	L&SGI (Scale 0 - 3)	Pooled 1.90 (0.20) Self Br 1.92 (0.16) Assisted Br 1.86 (0.23)	Pooled 1.90 (0.16) Self Br 1.88 (0.20) Assisted Br 1.96 (0.12)	56	USA	Pooled mean and SD calculated from the reported data. Cluster RCT.	There was no strong evidence of a difference in gingival indices between toothbrushes at 6 months.
6 months	Garcia-Carillo 2016	Sonicare EasyClean, Philips Electric TB® versus Vitis Access, Dentaids manual TB®	BDL&SGI (Scale 0-3)	1.24 (0.28)	1.19 (0.28)	64	Spain	The description of ID in this study appears to include some people with an IQ above 70.	There was no strong evidence of a difference in gingival indices between toothbrushes at 6 months. (P = 0.37). Pg. 5.
12 months	Carr 1997	Interplak Electric TB® versus an Oral B 40 manual TB®.	L&SGI (Scale 0 - 3)	Pooled 1.25 (0.47) Self Br 1.72 (0.30) Assisted Br 1.72 (0.42)	Pooled 1.73 (0.61) Self Br 1.87 (0.21) Assisted Br 2.0 (0.0)	56	USA	Pooled mean and SD calculated from the reported data. Cluster RCT.	RM analysis of variance showed the GI at 12 M was sig. associated with the toothbrush used (P = 0.017) positive for the Interplak Electric TB.

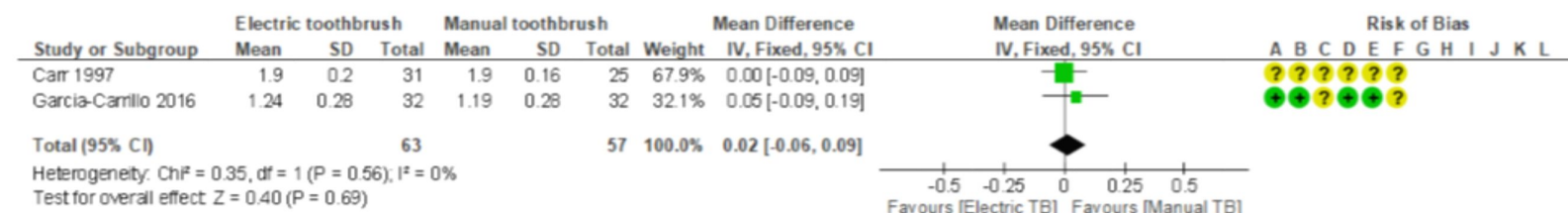
Footnotes

Kelner 1963 Subjective data presented. Data also presented for frequency of brushing. Carr 1997; Means and SDs were presented separately for self-brushing and assisted brushing, the pooled means and SDs have been calculated from these data for the different time points.

Indices: L&SGI: Loe & Silness Gingival Index, Scale 0 - 3. BDL&SGI: Bentley & Disney (four sites per tooth in two randomly (by coin toss) selected quadrants (one in the upper jaw, one in the lower jaw, contralateral); Scale 0-3. SGR; Subjective Gingival Rating, Scale Worse, Same, Better.

2 Electric toothbrush versus manual toothbrush for people with intellectual disabilities

2.1 Gingival inflammation medium term (6 weeks to 12 months)



Risk of bias legend

- (A) Random sequence generation (selection bias)
- (B) Allocation concealment (selection bias)
- (C) Blinding of participants and personnel (performance bias)
- (D) Blinding of outcome assessment (detection bias)
- (E) Incomplete outcome data (attrition bias)
- (F) Selective reporting (reporting bias)
- (G) Was the intervention independent of other changes? (ITS)
- (H) Was the shape of the intervention effect pre-specified? (ITS)
- (I) Was the intervention unlikely to affect data collection? (ITS)
- (J) Was knowledge of the allocated interventions adequately prevented during the study? (ITS)
- (K) Were incomplete outcome data adequately addressed? (ITS)
- (L) Was the study free from selective outcome reporting? (ITS)

Figure 13: C2 Meta-analysis Gingival inflammation - medium term

Table 27: C2 NRS Gingival inflammation studies - < 12 weeks

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
8/10 weeks	Gertenrich 1967a	"Oral B Automatic Toothbrush - Arcuate actions [®] " versus a manual brush (Py-co-pay, Hard, Lactona Jr. [®])	SGR (Scale 1-3)	Pooled 57.5% Group 1; 45%, Group 2; 70%	Pooled 77% Group 1; 54%, Group 2; 100%	70	USA	Data were presented as mean level of "obvious gingivitis" by % in a graph, Pooled % for the reported data were calculated.	General reduction in gingivitis with both brushes. A greater reduction in Group 1 (Participants with Down syndrome) for both brushes.
8 weeks	Gertenrich 1967b	"Oral B Automatic Toothbrush - Arcuate actions [®] " versus a manual brush (Py-co-pay, Hard, Lactona Jr. [®])	SGR (Scale 1-3)	Pooled 34% Group 1; 10%, Group 2; 58%	Pooled 43% Group 1; 50%, Group 2; 36%	37	USA	Data were presented as mean level of "obvious gingivitis" by % in a graph, Pooled % for the reported data were calculated.	General reduction in gingivitis in both groups and with both brushes. The greatest reduction was in Group 1 using the Electric brush.
8 weeks	Gertenrich 1967c	"Oral B Automatic Toothbrush - Arcuate actions [®] " versus a manual brush (Py-co-pay, Hard, Lactona Jr. [®])	SGR (Scale 1-3)	Pooled 83% Group 1; 74%, Group 2a; 85%, Group 2b; 100%	Pooled 54% Group 1; 50%, Group 2c; 50%, Group 2d; 65%	38	USA	Data were presented as mean level of "obvious gingivitis" by % in a graph, Pooled % for the reported data were calculated.	There was a general reduction in both groups and both brushes with a tendency for greater reduction in those using a standard brush.
8 weeks	Gertenrich 1967d	"Oral B Automatic Toothbrush - Arcuate actions [®] " versus a manual brush (Py-co-pay, Hard, Lactona Jr.)	SGR (Scale 1-3)	84%	74%	43	USA	Data were presented as mean level of "obvious gingivitis" by % in a graph.	A general reduction with no difference between brushes.
12 weeks	Bratel 1991	Braun 3D Electric toothbrush [®] versus Butler 411 manual toothbrush [®] .	L&SGI (Scale 0 -3)	Pooled 1.65 (0.10) Group 1; 1.6 (0.1) Group 2; 1.7 (0.1)	Pooled 1.60 (0.16) Group 1; 1.6 (0.2), Group 2; 1.6 (0.1)	23	Sweden	Pooled mean and SD calculated from the reported data.	No strong evidence of a difference in GI between the two brushes. Group 1 (unaided) using the electric TB brush was the only sub group with strong evidence of improvement from baseline at 12 weeks (P < 0.01) Table 2 Pg. 25.

Footnotes

Gertenrich 1967a; Data were presented as % difference separately for two distinct ID groups (Group 1: Down syndrome, Group 2: IQ of 20 or <) the pooled % differences have been calculated from these data. All subjects' teeth were brushed by attendants. Gertenrich 1967b; Data were presented as % difference separately for two distinct groups with ID (Group 1: Hydrocephaly and seizure unit, Group 2: Pediatric unit), the pooled % differences have been calculated from these data. All subjects' teeth were brushed by attendants. Gertenrich 1967c; In Group 1 (Cerebral palsied patients with IQ 30 - 95) all were brushed by attendants. In Group 2 ("Trainable patients" with IQ 30 - 50) half the subjects were brushed by attendants (2a and 2c) the other half self-brushed (2b and 2d). Gertenrich 1967d; All subjects were self-brushers with supervision and assistance from the attendants. Bratel 1991; Mean and SD presented for unaided brushing (Group 1), aided brushing (Group 2) and pooled mean and SDs have been calculated from these data.

Indices: SGR -Subjective Gingival Rating, measure of "Periodontal involvement"; Obvious signs of gingivitis" "Decrease in severity" and "Absence of clinical signs of gingivitis". "Obvious signs" and "decrease in severity" were combined on the graph. L&SGI - Loe & Silness Gingival Index; Scale 0-3.

Table 28: C2 NRS Gingival inflammation studies - >12 weeks

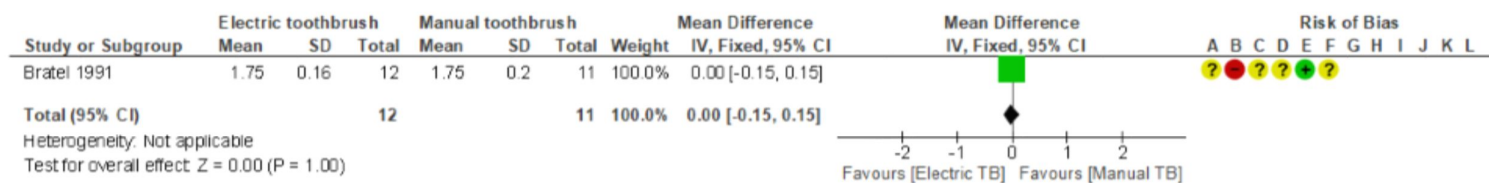
Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
21/28 weeks	Gertenrich 1967a	"Oral B Automatic Toothbrush - Arcuate actions [®] " versus a manual brush (Py-co-pay, Hard, Lactona Jr. [®])	SGR (Scale 1-3)	Pooled 53.5% Group 1; 37%, Group 2; 70%	Pooled 85% Group 1; 70%, Group 2; 100%	72	USA	Data were presented as mean level of "obvious gingivitis" by % in a graph, Pooled % for the reported data were calculated.	The reduction in obvious gingivitis seen at 11 weeks was maintained for both Group 1 (Down syndrome) and Group 2 (IQ < 20) using the Electric TB at this time point. Both groups using the manual TB regressed to baseline levels.
34 weeks	Gertenrich 1967b	"Oral B Automatic Toothbrush - Arcuate actions [®] " versus a manual brush (Py-co-pay, Hard, Lactona Jr. [®])	SGR (Scale 1-3)	Pooled 42.5% Group 1; 40%, Group 2; 45%	Pooled 40% Group 1; 30%, Group 2; 50%	36	USA	Data were presented as mean level of "obvious gingivitis" by % in a graph, Pooled % for the reported data were calculated.	Some reduction in gingivitis was maintained from baseline in both groups. No difference was found between the two brushes.
20 weeks	Gertenrich 1967c	"Oral B Automatic Toothbrush - Arcuate actions [®] " versus a manual brush (Py-co-pay, Hard, Lactona Jr. [®])	SGR (Scale 1-3)	Pooled 100% Group 1; 100%, Group 2a; 100%, Group 2b; 100%	Pooled 75% Group 1; 60%, Group 2a; 85%, Group 2b; 85%	36	USA	Data were presented as mean level of "obvious gingivitis" by % in a graph, Pooled % for the reported data were calculated.	Levels of observable gingivitis regressed towards baseline levels, with slightly less regression in the those using the Standard toothbrush.
20 weeks	Gertenrich 1967d	"Oral B Automatic Toothbrush - Arcuate actions [®] " versus a manual brush (Py-co-pay, Hard, Lactona Jr. [®])	SGR (Scale 1-3)	100%	100%	42	USA	Data were presented as mean level of "obvious gingivitis" by % in a graph.	Regression to baseline levels of gingivitis in both groups, although a "decrease in severity" was noted. There was no difference between the brushes.
16 months	Bratel 1991	Braun 3D Electric toothbrush [®] versus Butler 411 manual toothbrush [®] .	L&SGI (Scale 0-3)	Pooled 1.75 (0.16) Group 1; 1.7 (0.1), Group 2; 1.8 (0.2)	Pooled 1.70 (0.20) Group 1; 1.7 (0.2), Group 2; 1.7 (0.2)	23	Sweden	Data presented for aided and unaided with SD, overall pooled mean and SD calculated from the reported data.	No strong evidence of a difference in gingivitis between the brushes. The unaided electric TB sub group showed greater improvement at 16 months (p < 0.05). The aided manual TB sub group showed greater deterioration at 16 months (p < 0.01)

Footnotes

Gertenrich 1967a; Data were presented as % difference separately for two distinct ID groups (Group 1: Down syndrome, Group 2: IQ of 20 or <), the pooled % differences have been calculated from these data. All subjects' teeth were brushed by attendants. During the follow-up period Group 1 were told to expect regular oral hygiene checks, Group 2 were told the monitoring was over. Gertenrich 1967b; Data were presented as % difference separately for two distinct groups with ID (Group 1: Hydrocephaly and seizure unit, Group 2: Pediatric unit), the pooled % differences have been calculated from these data. All subjects' teeth were brushed by attendants. The findings of the study were discussed with the Group 2 attendants at week 20 which may have impacted on the results. Gertenrich 1967c; In Group 1 (Cerebral palsied patients with IQ 30 - 95) all were brushed by attendants. In Group 2 ("Trainable patients" with IQ 30 - 50) half the subjects were brushed by attendants (2a and 2c) the other half self-brushed (2b and 2d). Gertenrich 1967d; All subjects were self-brushers with supervision and assistance from the attendants. Bratel 1991; Mean and SD presented for unaided (Group 1) and aided (Group 2) brushing, pooled mean and SDs have been calculated from these data.

Indices: SGR -Subjective Gingival Rating, measure of "Periodontal involvement"; Obvious signs of gingivitis" "Decrease in severity" and "Absence of clinical signs of gingivitis". "Obvious signs" and "decrease in severity" were combined on the graph. L&SGI - Loe & Silness Gingival Index; Scale 0-3.

2.2 NRS Gingival inflammation long term (> 12 months)



Risk of bias legend

- (A) Random sequence generation (selection bias)
- (B) Allocation concealment (selection bias)
- (C) Blinding of participants and personnel (performance bias)
- (D) Blinding of outcome assessment (detection bias)
- (E) Incomplete outcome data (attrition bias)
- (F) Selective reporting (reporting bias)
- (G) Was the intervention independent of other changes? (ITS)
- (H) Was the shape of the intervention effect pre-specified? (ITS)
- (I) Was the intervention unlikely to affect data collection? (ITS)
- (J) Was knowledge of the allocated interventions adequately prevented during the study? (ITS)
- (K) Were incomplete outcome data adequately addressed? (ITS)
- (L) Was the study free from selective outcome reporting? (ITS)

Figure 14: C2 Meta-analysis NRS Gingival inflammation - long term

Table 29: C2 Plaque studies - short term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
1 week	Dögan 2004	Braun Plaque Control 3D Electric TB® versus Oral B Cross Action 35/40 manual TB®	MQHPI (Scale 0-5)	1.54 (0.71)	2.15 (0.58)	15	Turkey	Cross-over trial. Data are presented for the 2 arms separately at the final examination. Participant number at baseline unclear: assumed to be the same as at final time point.	There was strong evidence of a difference in the PI between the Braun 3D electric TB and Oral B Cross Action TB ($p < 0.001$) in favour of the electric TB.
2 weeks	Christen 2007	Dentacare-Sonodont Electric TB® versus Oral B Manual TB®	MQHPI (Scale 0-5)	Pooled Median 25.8 Self Br 23.0 Assist Br 27.5 Other Br 27.0	Pooled Median 28.3 Self Br 26.0 Assist Br 28.0 Other Br 31.0	72	Germany	Cross-over trial. Data are presented for the 2 arms combined. Pooled median data calculated from reported data	There was no strong evidence of a difference in the QH median across all patients between the three different toothbrushes in the final examination (Friedman test: $p > 0.05$, Tab. 13). Pg. 55.
2 weeks	Kaschke 2005	Teledyne Waterpik Sonic Speed Electric TB® versus Oral B Manual TB®	MQHPI (Scale 0-5)	Whole Mouth 3.1 (0.09) Surface Score 36.1(1.2)	Whole Mouth 3.25 (0.10) Surface Score 38.9 (1.9)	54	Germany	Cross-over trial. Data are presented for the 2 arms combined. Surface score Mean and SD obtained from author. Data calculated from tooth surface scores. Median presented in published article.	There was strong evidence that the mean plaque index for the Teledyne Waterpik was lower than the Oral B manual toothbrush ($p < 0.05$). Pg. 68.
4 weeks	Shaw 1983	Braun Electric TB® versus Oral B 30 manual TB®	S&LPI (Scale 0-3)	Total Mouth 0.59 (0.26) Surface Score 14.1 (6.2)	Total Mouth 0.57 (0.2) Surface Score 13.6 (4.7)	106	UK	Cross-over Trial. Data are presented for the two arms combined. Data calculated from tooth surface scores.	No strong evidence of a difference in oral hygiene performance when comparing the use of electric and manual toothbrushes. "There was no sig. diff between the groups ($t=0.833$, d.f.52, $p > 0.05$).". Pg.5

Footnotes

Dögan 2004; Three-armed cross-over trial; also compared the Superbrush, reported separately. Data presented is for the final examination, number of participants has been adjusted accordingly. Data for age groups and surfaces also reported. Christen 2007; Three-armed cross-over trial; also compared the Superbrush, reported separately. Median and percentile data only reported. Data presented for self-brushers, assisted brushers and other brushers, pooled data have been calculated from these data. Data for buccal and lingual surfaces, maxilla and mandible also reported. Kaschke 2005; Three-armed cross-over trial; also compared the Superbrush, reported separately. Data for buccal and lingual surfaces, maxilla and mandible and self-brushers, assisted brushers and other brushers also reported. Shaw 1983; Two-armed cross-over trial. Data presented for tooth surfaces with SD, mean total mouth score and SD calculated.

Indices: MGHPI: Modified Quigley & Hein Plaque Index; Scale of 0 - 5. S&L PI: Silness & Loe Plaque Index; Scale 0-3.

Table 30: C2 Plaque studies - medium term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
3 months	Carr 1997	Interplak Electric TB® versus an Oral B 40 manual TB®.	SG&VDI (Scale 0 - 3)	Pooled 1.31 (0.57) Self Br 1.39 (0.42) Assisted Br 1.19 (0.68)	Pooled 1.21 (0.68) Self Br 1.14 (0.49) Assisted Br 1.39 (0.82)	56	USA	Pooled mean and SD calculated from reported data. Cluster RCT.	There was no strong evidence of a difference in plaque levels between the tooth brushes at 3 months.
3 months	Garcia-Carrillo 2016	Sonicare EasyClean, Philips Electric TB® versus Vitis Access, Dentaaid manual TB®.	BDS&LPI (Scale 0 - 3)	1.02 (0.33)	1.12 (0.33)	64	Spain	The description of ID in this study appears to include some people with an IQ above 70.	There was no strong evidence of a difference in plaque levels between the toothbrushes at 3 months.
4 months	Kelner 1963	Automated electric TB (Broxodent®) versus Conventional TB	SOHR	% Improvement 27%	% Improvement 18%	100	USA	% Improvement calculated from the reported data.	"The automatic electric toothbrush appeared significantly superior to the conventional toothbrush in improving oral hygiene"
6 months	Carr 1997	Interplak Electric TB® versus an Oral B 40 manual TB®.	SG&VDI (Scale 0 - 3)	Pooled 1.55 (0.67) Self Br 1.60 (0.53) Assisted 1.46 (0.78)	Pooled 1.34 (0.41) Self Br 1.07 (0.51) Assisted 2.03 (0.29)	56	USA	Pooled mean and SD calculated from reported data. Cluster RCT.	There was no strong evidence of a difference in plaque levels between the tooth brushes at 6 months.
6 months	Garcia-Carrillo 2016	Sonicare EasyClean, Philips Electric TB® versus Vitis Access, Dentaaid manual TB®.	BDS&LPI (Scale 0 - 3)	1.12 (0.35)	1.04 (0.35)	64	Spain	The description of ID in this study appears to include some people with an IQ above 70.	There was no strong evidence of a difference in plaque levels between toothbrushes at 6 months. (p = 0.87) Pg. 4. Table 3.
12 months	Carr 1997	Interplak Electric TB® versus an Oral B 40 manual TB®.	SG&VDI (Scale 0 - 3)	Pooled 1.35 (0.57) Self Br 1.25 (0.51) Assisted Br 1.15 (0.63)	Pooled 1.49 (0.51) Self Br 1.25 (0.42) Assisted Br 2.10 (0.58)	56	USA	Pooled mean and SD calculated from reported data. Cluster RCT.	No reported strong evidence of a difference in plaque levels between tooth brushes at 12 months.

Footnotes

Kelner 1963; Subjective data presented, % improvement has been calculated from the differences between the combined Fair and Good ratings pre and post. Data also presented by age groups (Ages 4 to 14, 15 to 32). Carr 1997; Means and SDs were presented separately for self-brushing and assisted brushing, the pooled means and SDs have been calculated from these data for the different time points. Some differences noted between self and assisted brushers. "There was a marginally significant difference in the pattern of debris index across time for the two brushing assistance status groups (p = 0.054)." Pg. 135. Garcia-Carrillo 2016 reported the mean IQ as 60.6, ranging between 44 and 87.

Indices: SG&VDI: Simplified Green & Vermillion Debris Index; Scale 0 - 3, BDS&LPI: Bentley & Disney Modified Silness & Loe Plaque Index; Scale 0 - 3. SOHR: Subjective Oral Hygiene Rating; Scale Poor, Fair, Good.

Study or Subgroup	Electric toothbrush			Manual toothbrush			Weight	Std. Mean Difference IV, Fixed, 95% CI	Std. Mean Difference IV, Fixed, 95% CI	Risk of Bias															
	Mean	SD	Total	Mean	SD	Total				A	B	C	D	E	F	G	H	I	J	K	L				
Garcia-Carrillo 2016	1.12	0.35	32	1.04	0.35	32	53.9%	0.23 [-0.27, 0.72]																	
Carr 1997	1.55	0.67	31	1.34	0.41	25	46.1%	0.36 [-0.17, 0.90]																	
Total (95% CI)			63				100.0%	0.29 [-0.07, 0.65]																	

Heterogeneity: Chi² = 0.14, df = 1 (P = 0.71); I² = 0%

Test for overall effect: Z = 1.57 (P = 0.12)

Favours [Electric TB] Favours [Manual TB]

- (A) Random sequence generation (selection bias)
- (B) Allocation concealment (selection bias)
- (C) Blinding of participants and personnel (performance bias)
- (D) Blinding of outcome assessment (detection bias)
- (E) Incomplete outcome data (attrition bias)
- (F) Selective reporting (reporting bias)
- (G) Was the intervention independent of other changes? (ITS)
- (H) Was the shape of the intervention effect pre-specified? (ITS)
- (I) Was the intervention unlikely to affect data collection? (ITS)
- (J) Was knowledge of the allocated interventions adequately prevented during the study? (ITS)
- (K) Were incomplete outcome data adequately addressed? (ITS)
- (L) Was the study free from selective outcome reporting? (ITS)

124

Table 31: C2 NRS Plaque studies - < 12 weeks

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
0-8/10 weeks	Gertenrich 1967a	"Oral B Automatic Toothbrush - Arcuate actions [®] " versus a manual brush (Py-co-pay, Hard, Lactona Jr. [®])	SOHR (Scale 1 - 4)	Pooled mean Diff 0.75 (0.64) Group 1; 0.63 (0.71), Group 2; 1.18 (0.56)	Pooled mean Diff 0.59 (0.91) Group 1; 0.32 (1.08), Group 2; 1.12 (0.70)	55	USA	Pooled mean Difference and SD calculated from the reported data.	The results showed that both groups (Elec V manual TB) reduced plaque levels during the supervised brushing period, with the Elec TB being "somewhat better".
0-8 weeks	Gertenrich 1967b	"Oral B Automatic Toothbrush - Arcuate actions [®] " versus a manual brush (Py-co-pay, Hard, Lactona Jr. [®])	SOHR (Scale 1 - 4)	Pooled mean Diff 1.26 (0.50) Group 1; 1.70 (0.47), Group 2; 0.66 (0.53)	Pooled mean Diff 1.45 (0.73) Group 1; 1.57 (0.68), Group 2; 1.25 (0.77)	37	USA	Pooled mean Difference and SD calculated from the reported data.	An improvement in OH in both ID groups and with both brushes. (p ranged from 0.05 - 0.001). There was no strong evidence of a difference in oral hygiene between the two brushes.
0-8 weeks	Gertenrich 1967c	"Oral B Automatic Toothbrush - Arcuate actions [®] " versus a manual brush (Py-co-pay, Hard, Lactona Jr. [®])	SOHR (Scale 1-4)	Group 1 mean Diff 2.21 (0.60)	Group 1 mean Diff 1.72 (0.55)	Group 1; 15	USA	Mean Difference and SD reported for Group 1. Mean Square Difference was reported for Group 2. Type of Brush - Mean Sq (F) 2.67 (10.35). Author reports raw data no longer available.	A reduction in plaque levels in both ID groups and with both brushes (p ranged from 0.05 - 0.001). There was no strong evidence of a difference between the two brushes.
0-8 weeks	Gertenrich 1967d	"Oral B Automatic Toothbrush - Arcuate actions [®] " versus a manual brush (Py-co-pay, Hard, Lactona Jr. [®])	SOHR (Scale 1-4)	Mean Diff 1.36 (0.88)	Mean Diff 1.33 (0.58)	43	USA	Mean Diff presented in this study.	There was an improvement in OH with both brushes (p = 0.001) but no strong evidence of a difference in oral hygiene between the two brushes.
12 weeks	Bratel 1991	Braun 3D Electric toothbrush [®] versus Butler 411 Manual toothbrush [®] .	S&LPI (Scale 0 -3)	Pooled 1.10 (0.14) Unaided 0.9 (0.3), Aided 1.3 (0.2)	Pooled 1.11 (0.30) Unaided 1.2 (0.3), Aided 1.0 (0.3)	23	Sweden	Pooled mean and SD calculated from the reported data.	Some strong evidence of a difference in subgroups (Manual aided (p < 0.01) and Electric unaided (p < 0.05), however, no strong overall difference in PI between toothbrushes.

Footnotes

Gertenrich 1967a; Means and SDs were presented separately for two distinct groups with ID (Group 1: Down syndrome, Group 2: IQ of 20 or <), the pooled mean and SD have been calculated from these data. All subjects teeth were brushed by attendants. "Although the results obtained using the automatic units in a controlled regime were more stable and generally somewhat better (Figure 1) compared to the results with the hand brushing on a controlled regime, it is clear that these added benefits can vanish soon after the controlled regime is discontinued." Pg. 148. Gertenrich 1967b; Means and SDs were presented separately for two distinct ID groups (Group 1: Hydrocephaly and seizure unit, Group 2: Pediatric unit) the pooled mean and SD have been calculated from these data. All subjects' teeth were brushed by attendants. Gertenrich 1967c; Two distinct groups with ID (Group 1: Cerebral palsied patients with IQ 30 - 95, Group 2 "trainable patients" with IQ 30 - 50). In Group 1 all were brushed by attendants. In Group 2 half the subjects were brushed by attendants the other half self-brushed. In Group 2 "The most significant improvement occurred in the group that was brushed by attendants using the automated toothbrush" Pg. 157. Gertenrich 1967d; All subjects were self-brushers with supervision and assistance from the attendants. Bratel 1991; Mean and SD presented for aided and unaided brushing, pooled mean and SDs have been calculated from these data.

Indices: SOHR - Subjective Oral Hygiene Rating (Good, Fair, Poor and Very Poor); Scale 1-4. S&LPI - Silness & Loe Plaque Index; Scale 0-3.

Table 32: C2 NRS Plaque studies - > 12 weeks

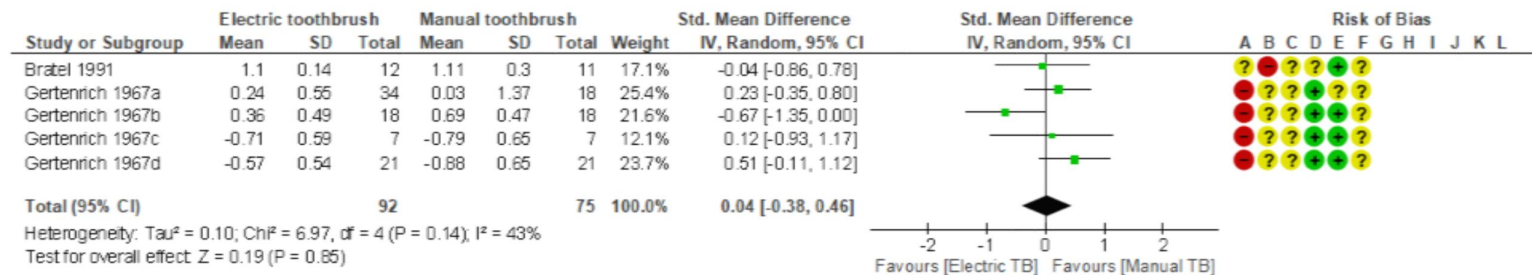
Duration	Study ID	Comparison	Index	Intervention Mean / SD	Control Mean / SD	Total No.	Country	Comments	Summary
8-20 weeks	Gertenrich 1967c	"Oral B Automatic Toothbrush - Arcuate actions®" versus a manual brush (Py-co-pay, Hard, Lactona Jr.®)	SOHR (Scale 1-4)	Group 1; -0.71 (0.59)	Group 1; -0.79 (0.65)	14	USA	Mean Difference and SD reported for Group 1. Mean Square Difference was reported for Group 2. Type of Brush - Mean Sq (F) 1.05 (1.54) NS. Author reports raw data no longer available.	There was no strong evidence of a difference in oral hygiene between the two brushes at this time point. "There was a significant regression in oral hygiene in both groups" at this time point. Pg. 158
8-20 weeks	Gertenrich 1967d	"Oral B Automatic Toothbrush - Arcuate actions®" versus a manual brush (Py-co-pay, Hard, Lactona Jr.®)	SOHR (Scale 1-4)	-0.57 (0.54)	-0.88 (0.65)	42	USA	Mean Difference presented in this study.	A slight regression in the improvements made in oral hygiene with both the Electric and Standard toothbrushes. (p= 0.001), there was no strong evidence of a difference in oral hygiene between the two brushes.
10 - 28 weeks	Gertenrich 1967a	"Oral B Automatic Toothbrush - Arcuate actions®" versus a manual brush (Py-co-pay, Hard, Lactona Jr.®)	SOHR (Scale 1-4)	Group 1; 0.24 (0.55)	Group 1; 0.03 (1.37)	52	USA	Mean Difference presented in this study for Group 1 only. Group 2 control joined Group 2 intervention after week 8.	There was no strong evidence of a difference in oral hygiene between the two brushes at this time point.
20 - 34 weeks	Gertenrich 1967b	"Oral B Automatic Toothbrush - Arcuate actions®" versus a manual brush (Py-co-pay, Hard, Lactona Jr.®)	SOHR (Scale 1-4)	Pooled 0.36 (0.49) Group 1; -0.10 (0.62), Group 2; 0.94 (0.30)	Pooled 0.69 (0.47) Group 1; 0.32 (0.61), Group 2; 1.28 (0.25)	36	USA	Pooled mean Difference and SD calculated from the reported data.	Strong evidence of an improvement in oral hygiene in Group 2 in both the Electric and standard toothbrushes (p = 0.05). No strong evidence of an improvement in either brush in Group 1.
16 months	Bratel 1991	Braun 3D Electric toothbrush® versus Butler 411 manual toothbrush®.	S&LPI (Scale 0-3)	Pooled 1.30 (0.35) Unaided 1.2 (0.3), Aided 1.4 (0.4)	Pooled 1.40 (0.30) Unaided 1.4 (0.3), Aided 1.4 (0.3)	23	Sweden	Pooled mean and SD calculated from the reported data.	"No significant changes concerning plaque index were found in or between groups after 16 months." Pg. 6.

Footnotes

Gertenrich 1967c; Means and SDs were presented separately for two distinct ID groups (Group 1: Cerebral palsied patients with IQ 30 - 95, Group 2 "trainable patients" with IQ 30 - 50), the pooled mean and SD have been calculated from these data. In Group 1 all were brushed by attendants. In Group 2 half the subjects were brushed by attendants the other half self-brushed. Gertenrich 1967d; Means and SDs were presented separately for two distinct ID groups, the pooled mean and SD have been calculated from these data. All subjects were self-brushers with supervision and assistance from the attendants. Gertenrich 1967a Data presented for only one sub group (Group 1: Down syndrome) as the control for the 2nd group were all using the Electric TB at this time point. Gertenrich 1967b; All subjects teeth were brushed by attendants. Data presented for two distinct groups with ID (Group 1: Hydrocephaly and seizure unit, Group 2: Pediatric unit). The findings of the study were discussed with the Group 2 attendants at week 20, which may have impacted on the results. Bratel 1991; Mean and SD presented for aided and unaided brushing, pooled mean and SDs have been calculated from these data.

Indices: SOHR - Subjective Oral Hygiene Rating (Good, Fair, Poor and Very Poor); Scale 1-4. S&LPI - Silness & Loe Plaque Index; Scale 0-3.

2.4 NRS Plaque Medium term (6 weeks to 12 months)



Risk of bias legend

- (A) Random sequence generation (selection bias)
- (B) Allocation concealment (selection bias)
- (C) Blinding of participants and personnel (performance bias)
- (D) Blinding of outcome assessment (detection bias)
- (E) Incomplete outcome data (attrition bias)
- (F) Selective reporting (reporting bias)
- (G) Was the intervention independent of other changes? (ITS)
- (H) Was the shape of the intervention effect pre-specified? (ITS)
- (I) Was the intervention unlikely to affect data collection? (ITS)
- (J) Was knowledge of the allocated interventions adequately prevented during the study? (ITS)
- (K) Were incomplete outcome data adequately addressed? (ITS)
- (L) Was the study free from selective outcome reporting? (ITS)

Figure 16: C2 Meta-analysis NRS Plaque - medium term

Table 33: C2 Calculus studies - medium term

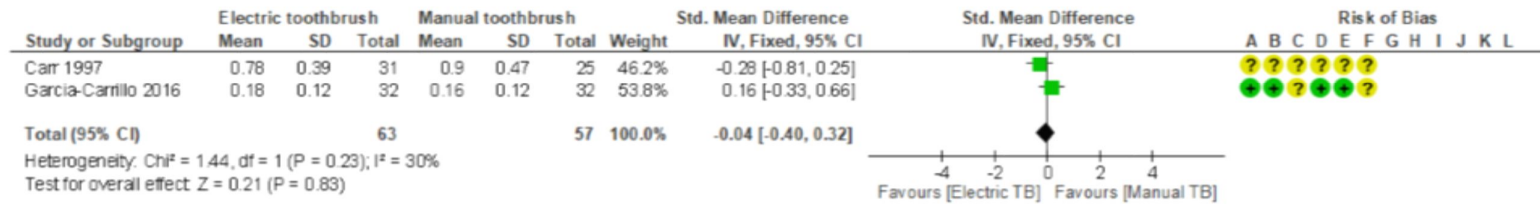
Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
4 months	Kelner 1963	Automated electric TB (Broxodent®) versus Conventional TB	SCR	% Improvement 13%	% Improvement 10%	100	USA	% Improvement calculated from the reported data.	"The automatic electric toothbrush appeared significantly superior to the conventional toothbrush in improving deposits" Pg. 108
6 months	Garcia-Carillo 2016	Sonicare EasyClean, Philips Electric TB® versus Vitis Access, Dentaïd manual TB®	BDCI (Scale 0 - 1)	0.18 (0.12)	0.16 (0.12)	64	Spain	The description of ID in this study appears to include some people with an IQ above 70.	No strong evidence of a difference in the effect of toothbrush group on the calculus index (p = 0.40)
6 months	Carr 1997	Interplak Electric TB® versus an Oral B 40 manual TB®.	SG&VCI (Scale 0 - 3)	Pooled 0.78 (0.39) Self Br 0.70 (0.41) Assisted Br 0.91 (0.37)	Pooled 0.90 (0.47) Self Br 0.77 (0.55) Assisted Br 1.23 (0.37)	56	USA	Pooled mean and SD calculated from the reported data. Cluster RCT	No evidence of a difference between groups could be found for the calculus index.
12 months	Carr 1997	Interplak Electric TB® versus an Oral B 40 manual TB®.	SG&VCI (Scale 0 - 3)	Pooled 0.76 (0.50) Self Br 1.81 (0.31) Assisted Br 1.78 (0.33)	Pooled 0.93 (0.40) Self Br 1.81 (0.31) Assisted Br 1.78 (0.33)	56	USA	Pooled mean and SD calculated from the reported data. Cluster RCT	No evidence of a difference between groups could be found for the calculus index.

Footnotes

Kelner 1963 Subjective data presented, the term Deposits was described as "the presence and amount of stain, soft and hard deposits" Pg. 102. % improvement has been calculated from the differences between the combined 'Extreme' ratings pre and post. Data also presented by age groups (Ages 4 to 14, 15 to 32). Carr 1997; Means and SDs were presented separately for self-brushing and assisted brushing, the pooled means and SDs have been calculated from these data for the different time points.

Indices: BDCI - Bentley & Disney Calculus Index; Present or absent, SG&VCI - Simplified Green & Vermillion Calculus Index; Scale 0-3. SCR; Subjective Calculus Rating, Scale None, Moderate, Extreme.

2.5 Calculus medium term (6weeks to 12 months)



Risk of bias legend

- (A) Random sequence generation (selection bias)
- (B) Allocation concealment (selection bias)
- (C) Blinding of participants and personnel (performance bias)
- (D) Blinding of outcome assessment (detection bias)
- (E) Incomplete outcome data (attrition bias)
- (F) Selective reporting (reporting bias)
- (G) Was the intervention independent of other changes? (ITS)
- (H) Was the shape of the intervention effect pre-specified? (ITS)
- (I) Was the intervention unlikely to affect data collection? (ITS)
- (J) Was knowledge of the allocated interventions adequately prevented during the study? (ITS)
- (K) Were incomplete outcome data adequately addressed? (ITS)
- (L) Was the study free from selective outcome reporting? (ITS)

Figure 17: C2 Meta-analysis Calculus - medium term

Table 34: C2 All outcomes and subgroups effect estimates

2 Electric toothbrush versus manual toothbrush for people with intellectual disabilities				
Outcome or Subgroup	Studies	Participants	Statistical Method	Effect Estimate
2.1 Gingival inflammation medium term (6 weeks to 12 months)	2	120	Mean Difference (IV, Fixed, 95% CI)	0.02 [-0.06, 0.09]
2.2 NRS Gingival inflammation long term (> 12 months)	1	23	Mean Difference (IV, Fixed, 95% CI)	0.00 [-0.15, 0.15]
2.3 Plaque medium term (6 weeks to 12 months)	2	120	Std. Mean Difference (IV, Fixed, 95% CI)	0.29 [-0.07, 0.65]
2.4 NRS Plaque Medium term (6 weeks to 12 months)	5	167	Std. Mean Difference (IV, Random, 95% CI)	0.04 [-0.38, 0.46]
2.5 Calculus medium term (6weeks to 12 months)	2	120	Std. Mean Difference (IV, Fixed, 95% CI)	-0.04 [-0.40, 0.32]

Comparison 3: Oral hygiene care training versus no oral hygiene care training or a placebo training session for carers of people with ID

3.1 Gingival inflammation (medium term)

Two RCTs, with 99 participants, compared the effect of oral hygiene care training of carers with no training of carers, on the levels of gingival inflammation of the people with ID for whom they cared, over periods of eight weeks and six to nine months (Mac Giolla Phadraig 2015; Phlypo 2016), (Table 35). A meta-analysis of these two studies with high heterogeneity ($\text{Chi}^2 = 3.16$, $\text{df} = 1$ ($P = 0.08$); $I^2 = 68\%$) showed no evidence of a difference in the medium term (MD: -0.09, 95% CI: -0.63 to 0.45), (Figure 18). The certainty of the evidence is low.

3.2 Plaque (medium term)

Two RCTs, with 99 participants, compared the effect of oral hygiene care training of carers with no training of carers on the plaque levels of the people with ID for whom they cared, over periods of eight weeks and six to nine months (Mac Giolla Phadraig 2015; Phlypo 2016), (Table 36). A meta-analysis of these two studies showed little or no effect size between the groups in the medium term (MD: -0.07, 95% CI: -0.26 to 0.13), (Figure 19). The certainty of the evidence is low.

3.3 NRS Plaque (short and medium term)

One NRCT with 34 participants compared the effect of oral hygiene care training of carers with no training of carers on the plaque levels of the people with ID for whom they cared, over a period of 21 days (Lange 2000), (Table 37).

In this NRCT, the people with ID, whose carers had training with accountability, showed strong evidence of lower plaque levels compared to the group without accountability (SMD -1.89, 95% CI -2.12 to -1.66). When combining the trained carers, both with and without accountability and comparing these to the carers who received no training, the strength of the evidence was slightly lower but still strong (MD: -0.69, 95% CI: -0.97 to -0.41), (Figure 20). The certainty of the finding is low.

One interrupted time series study with 11 participants compared the effect of oral hygiene care training of carers with no training of carers, on plaque levels of the people with ID for whom they cared, over a period of five months (Glassman 2006). Training of carers in oral hygiene care for people with ID may have a positive impact on the plaque levels of the people with ID for whom they care in the medium term (five months) if the carers are supported and encouraged to apply the training in practice subsequently (Table 38). Meta-analysis was not possible for this study due to the format of the data presented. The certainty of the findings is very low.

3.4 and 3.5 Knowledge of carers (short and medium term)

Three RCTs, with 213 participants (156 of whom were in 35 clusters), compared the effects of oral hygiene care training for carers of people with intellectual disabilities, over periods of 90 minutes, eight weeks and six to 11 months, with the effects of no training for carers or a placebo training session, on their oral care knowledge (Gonzalez 2013; Mac Giolla Phadraig 2015; Phlypo 2016), (Table 39).

The short-term study showed no evidence of a difference on oral health knowledge between intervention and control (MD 0.59, 95% CI -0.80 to 1.98), (Figure 21). The certainty of the evidence of the evidence was moderate. The two studies in the medium time frame, which used the same assessment questionnaire and had no heterogeneity, were included in a meta-analysis and showed a moderate effect on oral health knowledge in favour of training carers (MD: 0.69, 95% CI: 0.31 to 1.06), (Figure 22). The certainty of the evidence is determined to be low.

3.6 Behaviour of carers (short term/ < 6 weeks)

One RCT, with 193 carer participants, compared the effect of oral hygiene care training for carers with no training of carers, on behaviour only, over a period of four weeks, on five specific self-reported oral care behaviours (Olmos 2016), (Table 40).

The study reported strong evidence of positive differences for all five behaviours in the intervention from baseline ($P < 0.01$, no Mean or SD

provided, % data only), no strong evidence of positive differences was seen in the control group. The certainty of the evidence is moderate.

3.7 Behaviour, Attitude, Self-efficacy of carers (Medium term / 6 weeks - 12 months)

Two RCTs, one of which was a cluster RCT, with 189 carer participants compared the effect of oral hygiene care training for carers with no training of carers, on self-reported changes in Behaviour, Attitude and Self Efficacy together, over periods of eight weeks and six to 11 months (Mac Giolla Phadraig 2015; Phlypo 2016), (Table 41). Two studies, with moderate heterogeneity ($\text{Chi}^2 = 2.50$, $\text{df} = 1$ ($P = 0.11$); $I^2 = 60\%$), were included in a meta-analysis and showed no evidence of a difference, medium term (MD: 0.15, 95% CI: -0.80 to 1.10), (Figure 23). The certainty of the evidence is low.

Two interrupted time series studies with 15 carer participants examined the effect of oral hygiene care training of carers on their oral hygiene care skills and behaviour as well as the behaviour of the people with ID for whom they care (Glassman 2006; Kissel 1983), (Table 42). One of these studies reported general increases in carers use of training, a reduction in the level of assistance required and an "upward trend" in the level of self-initiated steps in the toothbrushing routine for the people with ID (Kissel 1983). The second study reported an increase in the caregivers presence during the toothbrushing session, an increase in the seconds spent brushing by the people with ID and a very slight increase in the aptitude of the person with ID in relation to toothbrushing (Glassman 2006). Meta-analysis was not possible for these studies due to the format of the data presented. The certainty of the evidence is determined to be very low.

The effect estimates for all outcomes and subgroups included in meta-analyses or Forest Plots for Comparison 3: Oral hygiene training for carers versus no oral hygiene training for carers, are presented in Table 43.

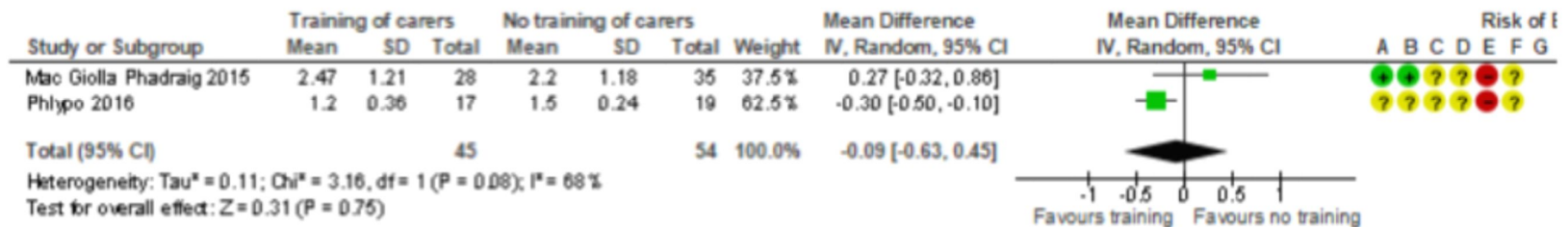
Table 35: C3 Gingival inflammation – medium term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
8 weeks	Phlypo 2016	Carers provided with an Information booklet and information session with practical skills.	MGI (Scale 0-3)	1.2 (0.36)	1.5 (0.24)	36	Belgium	Mean Diff data were reported; Intervention 0.1 (0.61); Control 0.003 (0.32)	No strong evidence of a difference was found in GI between the intervention and control group pre- and post-test ($P = 0.14$). However, when the individual post-test data was considered, a strong evidence of a difference ($P = 0.02$) in GI between the intervention and control group was found. Pg. 15, Table 2
6-9 months	Mac Giolla Phadraig 2015	Staff received training via a pyramidal training programme versus no training.	LMGI (Scale 0-4)	2.47 (1.21)	2.20 (1.18)	63	Ireland		"The difference in mean MGI and PI between groups was not statistically significant ($p > 0.05$, ANCOVA)." Pg. 95 "In this instance distal outcomes such as residents oral health and hygiene did not improve in the presence of proximal improvements in oral health-related knowledge, attitudes and self-efficacy, and reported behaviours of carers." Pg. 96

Footnotes

Indices: MGI: Modified Loe & Silness on buccal and lingual surfaces of 6 standard teeth, LMGI: Lobene Modified Gingival Index (non-invasive)

3.1 Gingival inflammation medium term (6 weeks to 12 months)



Risk of bias legend

- (A) Random sequence generation (selection bias)
- (B) Allocation concealment (selection bias)
- (C) Blinding of participants and personnel (performance bias)
- (D) Blinding of outcome assessment (detection bias)
- (E) Incomplete outcome data (attrition bias)
- (F) Selective reporting (reporting bias)
- (G) Was the intervention independent of other changes? (ITS)
- (H) Was the shape of the intervention effect pre-specified? (ITS)
- (I) Was the intervention unlikely to affect data collection? (ITS)
- (J) Was knowledge of the allocated interventions adequately prevented during the study? (ITS)
- (K) Were incomplete outcome data adequately addressed? (ITS)
- (L) Was the study free from selective outcome reporting? (ITS)

Figure 18: C3 Meta-analysis Gingival inflammation – medium term

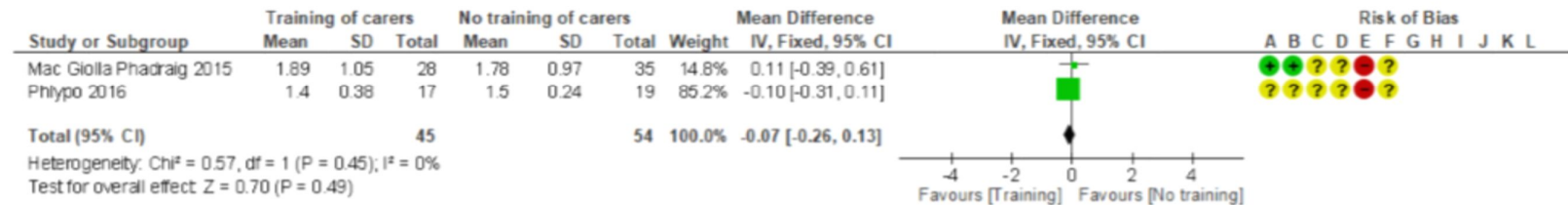
Table 36: C3 Plaque – medium term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
8 weeks	Phlypo 2016	Carers provided with an Information booklet and information session with practical skills.	MPI (Scale 0-3)	1.4 (0.38)	1.5 (0.24)	36	Belgium	Mean Diff also reported (p = 0.50)	No strong evidence of a difference in MPI. (p = 0.35) Pg. 15, Table 2
6-9 months	Mac Giolla Phadraig 2015	Staff received training via a pyramidal training programme on oral health and oral hygiene versus no training.	MPI (Scale 0-3)	1.89 (1.05)	1.78 (0.97)	63	Ireland	Brushing was carried out equally by the people with ID and the staff.	"The difference in mean MGI and PI between groups was not statistically significant (p > 0.05, ANCOVA)."Pg. 95

Footnotes

Indices: MPI: Modified Silness & Loe Plaque Index (6 standard teeth, no probe used, surfaces visible to the researcher).

3.2 Plaque medium term (6 weeks to 12 months)



Risk of bias legend

- (A) Random sequence generation (selection bias)
- (B) Allocation concealment (selection bias)
- (C) Blinding of participants and personnel (performance bias)
- (D) Blinding of outcome assessment (detection bias)
- (E) Incomplete outcome data (attrition bias)
- (F) Selective reporting (reporting bias)
- (G) Was the intervention independent of other changes? (ITS)
- (H) Was the shape of the intervention effect pre-specified? (ITS)
- (I) Was the intervention unlikely to affect data collection? (ITS)
- (J) Was knowledge of the allocated interventions adequately prevented during the study? (ITS)
- (K) Were incomplete outcome data adequately addressed? (ITS)
- (L) Was the study free from selective outcome reporting? (ITS)

Figure 19: C3 Meta-analysis Plaque – medium term

Table 37: C3 NRS Plaque – short term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
21 days	Lange 2000	Training of carers with and without accountability (G1 + G2) versus no training (G3)	MRPDI (Scale 0-3)	Pooled Data 1.09 (0.26). G1: 0.23 (0.02) G2: 2.12 (0.37)	1.78 (0.45)	34	USA	Pooled mean and SD calculated from reported data for G1 and G2.	The plaque index for Group 1 (with accountability) was lower than that of Group 2 (without accountability) (p=.004) and the control group (p= .0001).
21 days	Lange 2000	Training of carers with accountability (G1) versus training without (G2) accountability.	MRPDI (Scale 0-3)	0.23 (0.02)	2.12 (0.37)	24	USA	SD calculated from reported data for G1 and G2.	"Fisher's LSD indicted significantly lower plaque indexes for experimental Group I versus Group 2 (p=.00001)." Pg 208

Footnotes

Lange 2000; Mean and SE were presented separately for "with accountability" (G1) and "without accountability" (G2) of trainers, and for Control (G3), SDs were calculated from these data, mean and SD were pooled for G1 and G2 in Comparison 1 and compared in Comparison 2.

Indices: MRPDI -Modified Ramfjord's Periodontal Disease index -plaque only.

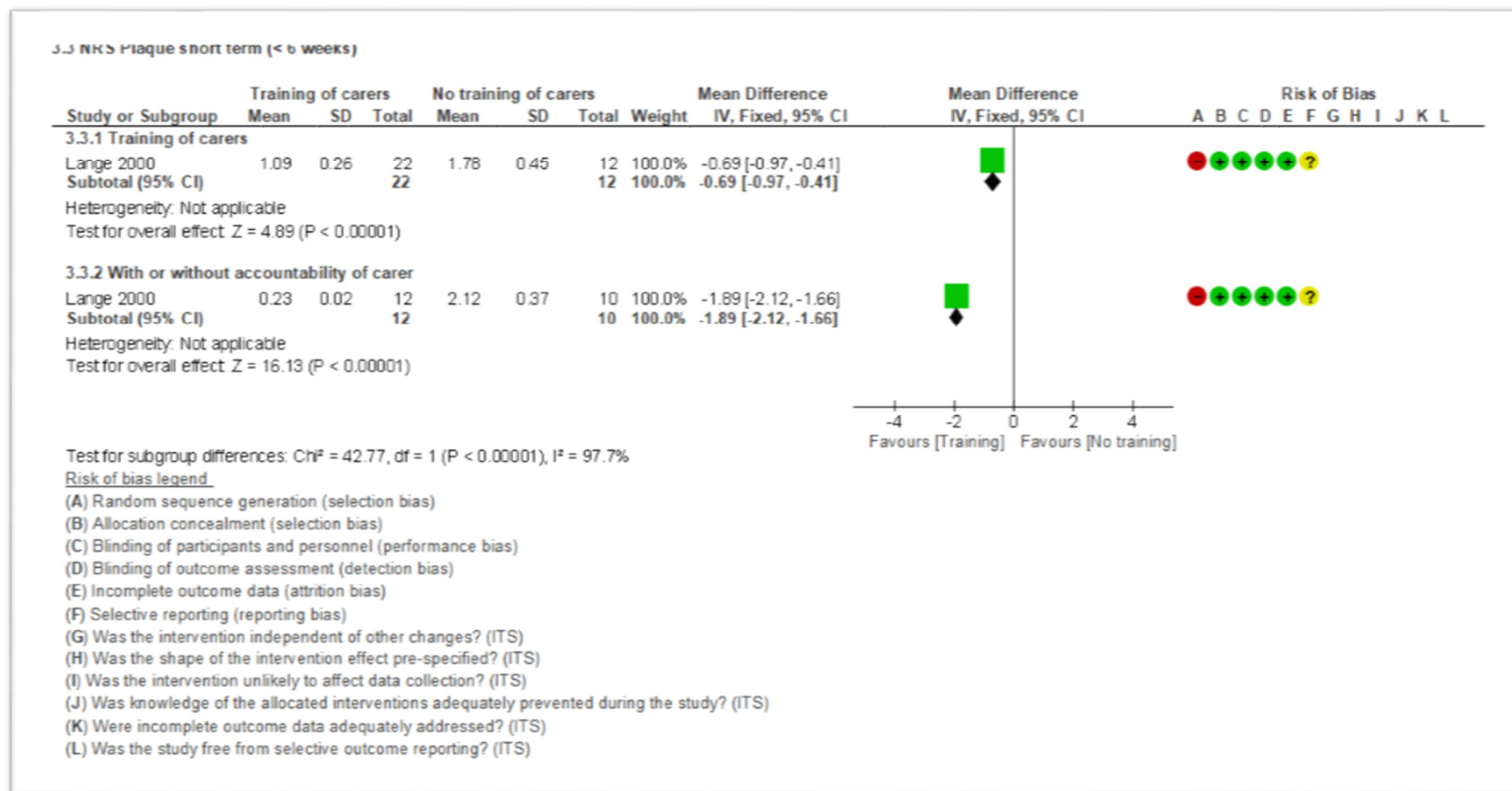


Figure 20: C3 Meta-analysis NRS Plaque – short term

Table 38: C3 NRS Plaque – medium term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
4-5 months	Glassman 2006	Effect of knowledge acquisition on carers behaviour and their client's oral hygiene behaviour.	SOHI (Scale 0-3)	Range 1.6 to 0.3	N/A	11	USA	Data in published article was unclear. Multiple attempts to contact authors failed.	"Improvement from baseline through the coaching phase of from 18.89% to 72.5%" Pg 43.

Footnotes

Indices: SOHI - Simplified Green & Vermillion Oral Hygiene Index, debris only; Scale 0 - 3

Table 39: C3 Knowledge – short and medium term

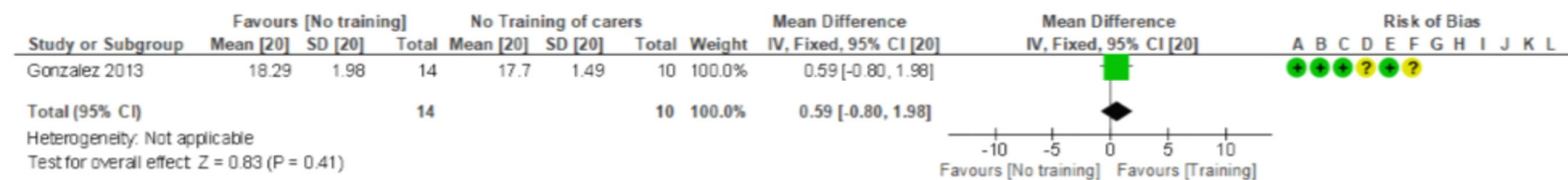
Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
90 mins	Gonzalez 2013	Oral Health lecture with hands on training (90 mins) versus an oral health discussion (30 mins).	Knowledge and comprehension (Scale 0 - 1 for 20 questions)	18.29 (1.98) Mean Diff 0.061 (0.086)	17.7 (1.49) Mean Diff 0.035 (0.094)	24	USA	Mean Difference is reported. Assessment was immediately pre and post intervention.	There was strong evidence of a difference between the two groups (P = 0.01). Both groups did improve their knowledge from baseline.
8 weeks	Phlypo 2016	Carers provided with an Information booklet and information session with practical skills.	Knowledge (Scale 0- 1 for 10 questions)	6.8 (1.43)	5.9 (1.33)	34	Belgium	Subgroup analyses were reported.	Post-test, overall no strong evidence of differences was reported in K-index scores between and within groups. Pg. 6.
6.5 - 11 months	Mac Giolla Phadraig 2015	Staff received training via a pyramidal training programme versus no training.	Knowledge (Scale 0 -1 for 10 question)	7.86 (1.27)	7.21 (1.32)	155	Ireland	Cluster RCT.	Strong evidence of a difference in favour of the intervention was reported (P = 0.002).

Footnotes

Gonzalez 2013; "Both groups show an increase in scores, however, the experimental group shows almost twice the increase as the control group (0.061 versus 0.035). The standard deviation of both of these groups is fairly large (around 0.09)." Pg. 296. Mean and SD calculated from individual data provided (Table 2, Pg. 296). Phlypo 2016; Subgroup analysis of those in the intervention group who read the information booklet compared to those in the control group who did not, showed strong evidence of an improvement in knowledge (p = 0.05). Mac Giolla Phadraig 2015; "Using independent-samples t-test, a significant difference between control and intervention is noted for K Index" (p = 0.002) Pg. 188 and Table 6 Pg. 189. The level of significance was adjusted to p = 0.026, using the rough false discovery rate.

Indices: Multiple choice questionnaires designed specifically for the studies for Gonzalez 2013 and Mac Giolla Phadraig 2015. Phlypo 2016 used the questionnaire used in the Mac Giolla Phadraig study.

3.4 Knowledge short term (< 6 weeks) [20]



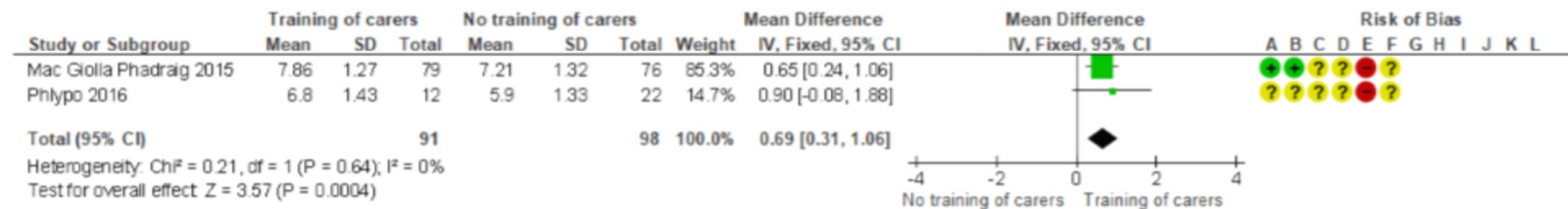
Risk of bias legend

- (A) Random sequence generation (selection bias)
- (B) Allocation concealment (selection bias)
- (C) Blinding of participants and personnel (performance bias)
- (D) Blinding of outcome assessment (detection bias)
- (E) Incomplete outcome data (attrition bias)
- (F) Selective reporting (reporting bias)
- (G) Was the intervention independent of other changes? (ITS)
- (H) Was the shape of the intervention effect pre-specified? (ITS)
- (I) Was the intervention unlikely to affect data collection? (ITS)
- (J) Was knowledge of the allocated interventions adequately prevented during the study? (ITS)
- (K) Were incomplete outcome data adequately addressed? (ITS)
- (L) Was the study free from selective outcome reporting? (ITS)

Figure 21: C3 Meta-analysis Knowledge – short term

3 Training of carers versus no training of carers

3.5 Knowledge medium term (6 weeks to 12 months)



Risk of bias legend

- (A) Random sequence generation (selection bias)
- (B) Allocation concealment (selection bias)
- (C) Blinding of participants and personnel (performance bias)
- (D) Blinding of outcome assessment (detection bias)
- (E) Incomplete outcome data (attrition bias)
- (F) Selective reporting (reporting bias)
- (G) Was the intervention independent of other changes? (ITS)
- (H) Was the shape of the intervention effect pre-specified? (ITS)
- (I) Was the intervention unlikely to affect data collection? (ITS)
- (J) Was knowledge of the allocated interventions adequately prevented during the study? (ITS)
- (K) Were incomplete outcome data adequately addressed? (ITS)
- (L) Was the study free from selective outcome reporting? (ITS)

Figure 22: C3 Meta-analysis Knowledge – medium term

Table 40: C3 Behaviour – short term

Duration	Study ID	Comparison	Index	Intervention %	Control %	Total No.	Country	Comments	Summary
4 weeks	Olmos 2016	Training of carers versus no training of carers	Pre and Post survey of carers	1. 61.10% 2. 70.40% 3. 54.10% 4. 78.60% 5. 98.90%	1. 25.50% 2. 17.90% 3. 2.10% 4. 29.50% 5. 64.20%	193	Germany	The three-headed toothbrush (SuperBrush) was used in this study.	Strong evidence of a difference in self-reported behaviour from baseline in the intervention group ($p < 0.01$)

Footnotes

Olmos 2016; Changes in Behaviour assessed: 1. Use of a special toothbrush, 2. Use of fluoride toothpaste, 3. Application of fluoride weekly, 4. Toothbrushing after breakfast and 5. Duration of toothbrushing

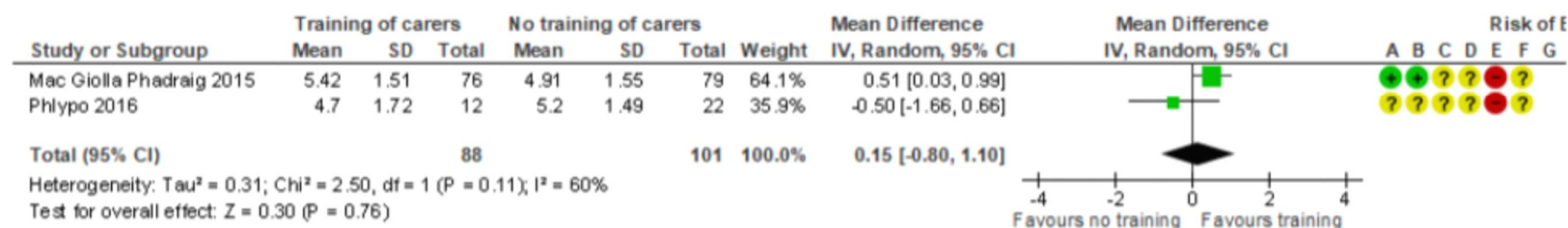
Table 41: C3 Behaviour, attitude and self-efficacy – medium term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
8 weeks	Phlypo 2016	Carers provided with an Information booklet and information session with practical skills.	BAS (14 questions)	4.7 (1.72)	5.2 (1.49)	34	Belgium	Subgroup analyses was reported on those who read the information booklet	Post-test, overall no strong evidence of differences was found in BAS-scores between and within groups. Pg. 6. ($p=0.38$)
8 - 11 months	Mac Giolla Phadraig 2015	Staff received training via a pyramidal training programme versus no training.	BAS (14 questions)	5.42 (1.51)	4.9 (1.55)	155	Ireland	The level of significance is adjusted to $p = 0.028$, using the rough false discovery rate.	"there was no difference in BAS scale scores ($p = 0.040$) between control and intervention at post-test." Pg. 188

Footnotes

Indices: BAS; Behaviour (-2 or +2) 4 questions, Attitude (-2, -1, 1, 2) 5 questions, Self-efficacy (-2, -1, 1, 2) 5 questions.

3.6 Behaviour, attitude and self-efficacy medium term



Risk of bias legend

- (A) Random sequence generation (selection bias)
- (B) Allocation concealment (selection bias)
- (C) Blinding of participants and personnel (performance bias)
- (D) Blinding of outcome assessment (detection bias)
- (E) Incomplete outcome data (attrition bias)
- (F) Selective reporting (reporting bias)
- (G) Was the intervention independent of other changes? (ITS)
- (H) Was the shape of the intervention effect pre-specified? (ITS)
- (I) Was the intervention unlikely to affect data collection? (ITS)
- (J) Was knowledge of the allocated interventions adequately prevented during the study? (ITS)
- (K) Were incomplete outcome data adequately addressed? (ITS)
- (L) Was the study free from selective outcome reporting? (ITS)

Figure 23: C3 Meta-analysis Behaviour, attitude and self-efficacy – medium term

Table 42: C3 NRS Behaviour and self-efficacy – medium term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
100 days	Kissel 1983	Training of carers to assist PID to brush their teeth using verbal instruction, physical guidance, rewards, and self-management procedures.	Carers use of training	Mean Diff 44% on average (Increase)	N/A	4	USA	% Mean Differences reported narratively. Two other life skills were also taught to other residents by the staff in this study.	General increases in correct use of all assistance techniques during training; physical assistance, verbal instruction and rewards. All staff used all types of assistance correctly on average >94% during the maintenance phase. Pg 404
100 days	Kissel 1983	Training of carers to assist PID to brush their teeth using verbal instruction, physical guidance, rewards, and self-management procedures.	PID's need for levels of assistance when tooth brushing	Mean Diff. Physical Assistance 15.5% (Reduction) Self-Initiation "Upward trend"	N/A	4	USA	Two other life skills were also taught to other residents by the staff in this study.	Need for physical assistance - Baseline 47% – 60%, reduced to 38% at end of training. Verbal Instruction was inconsistent at end of training. 'Upward trends' in self-initiated behaviour (varied by resident). Pg. 404
4-5 months	Glassman 2006	Effect of knowledge acquisition on carers behaviour.	Caregivers Presence (+/-) during the brushing session	Mean Diff 37 - 89% (increase)	N/A	11	USA	Data in Figure 1 presented unclearly in published article. Multiple attempts to contact authors failed. Data presented as a mean for each phase of the training (classroom sessions and coaching sessions) in the text.	"The phase mean for caregiver presence during oral hygiene sessions was 100% for all clients after the coaching phase." Pg. 42
4-5 months	Glassman 2006	Duration of Brushing	Seconds spent brushing.	Mean Diff 484% (increase)	N/A	11	USA	Data presented in Figure 1 unclear in published article. Multiple attempts to contact authors failed.	Duration of brushing increased from a phase mean of 20.5-73.8 seconds at baseline to 57.9 - 215.4 seconds after coaching. Pg. 42
4-5 months	Glassman 2006	Carers perception of client's aptitudes	Social Validation Questionnaire (Scale 0-6)	Mean Diff ranged from 0.0 to 0.75	N/A	11	USA		"The average scores showed only a slight increase". Pg. 43

Footnotes

Glassman 2006; Carers received training in the classroom (training phase) and were also provided with ongoing coaching and feedback (coaching phase) during their oral hygiene care sessions with the people with ID for whom they cared.

Table 43: C3 All outcomes and subgroups effect estimates

3 Training of carers of people with intellectual disabilities versus no training of carers				
Outcome or Subgroup	Studies	Participants	Statistical Method	Effect Estimate
3.1 Gingival inflammation medium term (6 weeks to 12 months)	2	99	Mean Difference (IV, Random, 95% CI)	-0.09 [-0.63, 0.45]
3.2 Plaque medium term (6 weeks to 12 months)	2	99	Mean Difference (IV, Fixed, 95% CI)	-0.07 [-0.26, 0.13]
3.3 NRS Plaque short term (< 6 weeks)	1		Mean Difference (IV, Fixed, 95% CI)	Subtotals only
3.3.1 Training of carers	1	34	Mean Difference (IV, Fixed, 95% CI)	-0.69 [-0.97, -0.41]
3.3.2 With or without accountability of carer	1	22	Mean Difference (IV, Fixed, 95% CI)	-1.89 [-2.12, -1.66]
3.4 Knowledge short term (< 6 weeks) [20]	1		Mean Difference (IV, Fixed, 95% CI [20])	Subtotals only
3.5 Knowledge medium term (6 weeks to 12 months)	2	189	Mean Difference (IV, Fixed, 95% CI)	0.69 [0.31, 1.06]
3.6 Behaviour, attitude and self-efficacy medium term	2	189	Mean Difference (IV, Random, 95% CI)	0.15 [-0.80, 1.10]

Comparison 4: Oral hygiene training versus no oral hygiene training for people with ID

4.1 Gingival Inflammation (short term / < 6 weeks)

One RCT, with ten participants, compared the effects of oral hygiene training for people with ID with no training, on gingival inflammation, over a six-to-seven-week time period (Lange 1985), (Table 44). The effect size was small in favour of oral hygiene training of people with ID (MD: -0.28, 95% CI: -0.90 to 0.34), (Figure 25). The certainty of the evidence is low.

4.2 Plaque (short term/ < 6 weeks)

Two RCTs, with 36 participants, compared the effects of oral hygiene training for people with ID with no training on plaque levels over a five-to-seven-week time period (Albino 1979; Lange 1985), (Table 45). One study, which had data that could be entered on a Forest Plot, showed a moderate effect size on plaque levels in favour of oral hygiene training of people with ID short term (MD: -0.47, 95% CI: -0.92 to -0.02), (Figure 26). The certainty of the evidence was determined to be low. The other study had similar findings in favour of training.

4.3 NRCT Behaviour (medium and long term)

Four interrupted time series studies, with 59 participants, examined the effect of oral hygiene training for people with ID on their toothbrushing behaviour over periods of 100 days, 30 weeks and 19 months (Bouter 1979; Jarman 1983; Snell 1989). The remaining study was unclear in relation to the duration of the intervention (Abramson 1972), (Table 46). Oral hygiene training of people with ID may impact on their toothbrushing behaviour but it is unclear as to how this will impact on their oral health. Meta-analysis was not possible for this outcome due to the format of the data presented. The certainty of the evidence is very low.

The effect estimates for all outcomes and subgroups included in Forest Plots for Comparison 4: Oral hygiene training versus no oral hygiene training for people with intellectual disabilities, are presented in Table 47.

Table 44: C4 Gingival inflammation – short term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
6/7 weeks	Lange 1985	Self-modelling training in oral hygiene skills with daily use of plaque disclosing agents versus no training but daily plaque scoring.	L&SGI (Scale 0-3)	Pooled Data 0.98 (0.54)	Pooled Data 1.26 (0.46)	10	USA	Mean and SD calculated from individual means.	"The gingival indices [for those in treatment] improved from 12% to 50%." Pg 260 The results of the gingival index for the control group was variable, with slight improvement, which was attributed to daily brushing.

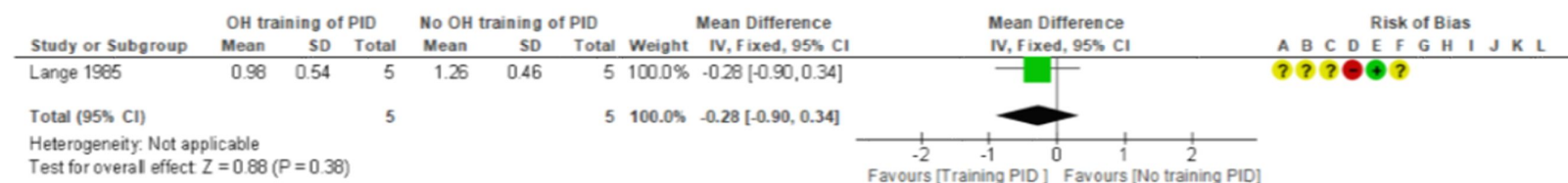
Footnotes

Lange 1985; data presented by individuals, pooled mean and SD calculated from these data.

Indices: S&LGI - Silness and Loe Gingival Index

4 Training of people with ID versus no training of people with ID

4.1 Gingival inflammation short term (< 6 weeks)



Risk of bias legend

- (A) Random sequence generation (selection bias)
- (B) Allocation concealment (selection bias)
- (C) Blinding of participants and personnel (performance bias)
- (D) Blinding of outcome assessment (detection bias)
- (E) Incomplete outcome data (attrition bias)
- (F) Selective reporting (reporting bias)
- (G) Was the intervention independent of other changes? (ITS)
- (H) Was the shape of the intervention effect pre-specified? (ITS)
- (I) Was the intervention unlikely to affect data collection? (ITS)
- (J) Was knowledge of the allocated interventions adequately prevented during the study? (ITS)
- (K) Were incomplete outcome data adequately addressed? (ITS)
- (L) Was the study free from selective outcome reporting? (ITS)

Figure 24: C4 Meta-analysis Gingival inflammation – short term

Table 45: C4 Plaque – short term

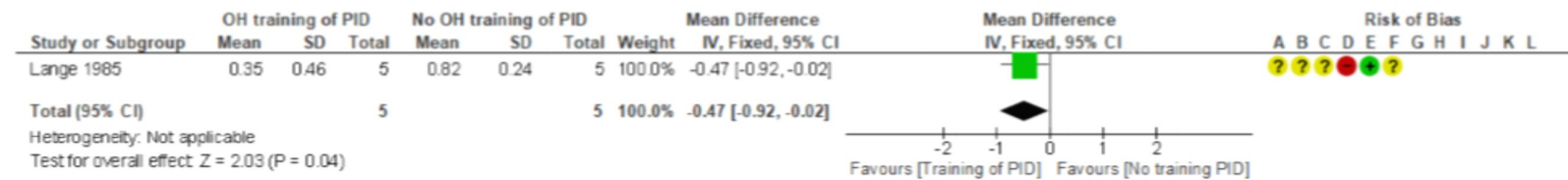
Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
5 weeks	Albino 1979	Desensitisation session and 11 practical training sessions in oral hygiene versus a desensitisation session only.	K&API (Scale 0-5)	2.31	3.04	26	USA	Data presented is the Adjusted Post Test scores. Some confusion re data between the two reports.	"indicating a statistically significant change for the experimental group compared to the control group" Schwartz Pg. 20 Analysis of covariance: F= 6.38; df = 1/23; p <.025. Albino Pg. 27
6/7 weeks	Lange 1985	Self-modelling training in oral hygiene skills with daily use of plaque disclosing agents versus no training but daily plaque scoring.	G&VPI (Scale 0-3)	Pooled Data 0.35 (0.46)	Pooled Data 0.82 (0.24)	10	USA	Mean and SD calculated from individual means.	The plaque level reduced for the intervention group from baseline to follow-up by 30%. Pg. 260. There was a general increase in plaque levels from baseline in the control group.

Footnotes

Lange 1985; data presented by individuals, pooled mean and SD calculated from these data.

Indices: K&API - Kobayashi & Ash Plaque Index (6 teeth scored, facial and lingual surfaces only). G&VPI - Green & Vermillion Plaque Index.

4.2 Plaque short term (< 6 weeks)



Risk of bias legend

- (A) Random sequence generation (selection bias)
- (B) Allocation concealment (selection bias)
- (C) Blinding of participants and personnel (performance bias)
- (D) Blinding of outcome assessment (detection bias)
- (E) Incomplete outcome data (attrition bias)
- (F) Selective reporting (reporting bias)
- (G) Was the intervention independent of other changes? (ITS)
- (H) Was the shape of the intervention effect pre-specified? (ITS)
- (I) Was the intervention unlikely to affect data collection? (ITS)
- (J) Was knowledge of the allocated interventions adequately prevented during the study? (ITS)
- (K) Were incomplete outcome data adequately addressed? (ITS)
- (L) Was the study free from selective outcome reporting? (ITS)

Figure 25: C4 Meta-analysis Plaque – short term

Table 46: C4 NRS Self-efficacy – medium and long term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
Unclear	Abramson 1972	OH training for people with ID using discrimination reinforcement and modelling	Steps in a TB routine (19)	Pooled data Mean Diff 6.3 steps (increase)	N/A	8	USA	Group mean and SD calculated from the reported data.	One-tailed t- tests for correlated data showed strong evidence of positive mean differences in toothbrushing behaviour (range $p < .005$ to $p < .0005$). Table 3 Pg. 8
100 days	Bouter 1979	Oral hygiene training for people with ID using assistance, feedback and duration training.	Steps in a TB routine (15)	62.5% (increase)	N/A	8	NL	Number of training and duration training sessions varied considerably between participants.	62.5% of participants learned to brush their teeth and continued to do so after 8-15 days of no training.
30 weeks	Jarman 1983	Oral hygiene training for people with ID using a token reinforcement programme with single and chained response contingency phases.	Completing the TB skill	60.2% - 71.9% of participants (increase)	N/A	40	USA	Individual performance criteria were determined at baseline. Five other life skills were also taught in this study.	Mean weekly performance of tooth brushing increased by 60.2% using single contingency reinforcement and by 71.9% using chained contingency reinforcement.
19 months	Snell 1989	Oral hygiene training for people with ID using verbal and physical prompts which were gradually faded.	Steps in a TB routine (17)	Difference 5 - 12 Steps (increase)	N/A	3	USA	Results reported narratively and are unclear.	"Training was not consistently followed by maintenance of effects. Although Sara's performance was durable, Jake and Frank did not maintain all their skills without receiving 'booster' training sessions." Pg. 224

Footnotes

Abramson 1972; Mean individual data reported (no SD reported), Group Mean and SD calculated from these data.

Table 47: C4 All outcomes with effect estimates

4 Training of people with ID versus no training of people with ID

Outcome or Subgroup	Studies	Participants	Statistical Method	Effect Estimate
4.1 Gingival inflammation short term (< 6 weeks)	1	10	Mean Difference (IV, Fixed, 95% CI)	-0.28 [-0.90, 0.34]
4.2 Plaque short term (< 6 weeks)	1	10	Mean Difference (IV, Fixed, 95% CI)	-0.47 [-0.92, -0.02]
4.3 NRS Self-efficacy	1	14	Mean Difference (IV, Fixed, 95% CI)	-6.30 [-7.72, -4.88]

Comparison 5: One-, three- and six-monthly dental recall intervals versus no dental recall for people with ID

One cluster-RCT, with 304 participants, compared the benefit of 1-, 3- and 6-monthly dental recalls with no dental recalls, for people with ID, on gingival inflammation, gingival bleeding, plaque, and calculus over periods of 3, 6, 12, 18 and 24 months (Shaw 1991), the findings at 24 months are reported here (Table 48; Table 49; Table 50, Table 51).

5.1 Gingival inflammation (long term)

The meta-analysis showed effect sizes for the Code 1 score (gingival pockets) were moderate in favour of regular dental recalls (One monthly: MD: -0.60, 95% CI: -0.97 to -0.23, Three monthly: MD: -0.50, 95% CI: -0.85 to -0.15, Six monthly: MD: -0.40, 95% CI: -0.76 to -0.04), (Figure 27).

5.2 Gingival bleeding (long term)

There was a small effect size, at 24 months, for gingival bleeding in favour of one and three monthly dental recall intervals and a small effect size in favour of no regular dental recall compared to 6 monthly dental recalls (One monthly: MD: -0.20, 95% CI: -0.86 to 0.46, Three monthly: MD: -0.10, 95% CI: -0.73 to 0.53, Six monthly: MD: 0.20, 95% CI: -0.38 to 0.78), (Figure 28).

5.3 Plaque (long term)

The meta- analysis of data showed the effect size for plaque levels at 24 months was large in favour of more frequent dental recall intervals (One monthly: MD: -0.70, 95% CI: -1.28 to -0.12, Three monthly: MD: -0.80, 95% CI: -1.40 to -0.20 and Six monthly: MD: -0.70, 95% CI: -1.22 to -0.18), (Figure 29).

5.4 Calculus (long term)

The effect size at 24 months for calculus levels was large in favour of regular dental recalls, the more regular the recall the larger the effect size (One monthly: MD: -2.00, 95% CI: -2.64 to -1.36, Three monthly: MD: -1.40, 95% CI: -2.04 to -0.76, Six monthly: MD: -0.10, 95% CI: -0.72 to 0.52), (Figure 30).

The certainty of the evidence for all outcomes was determined to be low.

The effect estimates for all outcomes and subgroups included in meta-analyses for Comparison 5: One-, three- and six-monthly dental recall intervals versus no dental recall for people with ID are presented in Table 52.

Table 48: C5 Gingival inflammation – long term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
24 months	Shaw 1991	One, three- and six-monthly dental recall versus no regular dental recall	WHOGPI (Scale 0 - 2)	R1 mth: 0.3 (0.8), R3 mth: 0.4 (0.7), R6 mth: 0.5 (0.9)	0.9 (1.4)	304	UK	All participants also had supervised daily toothbrushing.	Strong evidence of differences between the groups for Code 1 GPI at 3, 6, 12 and 24 months ($p < 0.01 - 0.05$).

Footnotes

Shaw 1991; Data also reported for 3, 6, 12 and 18 months, not reported here. Data are for "Mean number of sextants scored as Code 1". No strong evidence of a difference for Code 2, but the actual number of effected sextants was very low.

Indices: WHOGPI; World Health Organisation Gingival Pocket Index; Scale 0 -2 (0; 0 -3.5mm, 1; >3.5 to 5.5mm, 2; >5.5mm)

Table 49: C5 Gingival bleeding – long term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
24 months	Shaw 1991	One, three- and six-monthly dental recall versus no regular dental recall	WHOB I (Scale 0 -1)	R1 mth: 2.2 (1.9), R3 mth: 2.3 (1.9), R6 mth: 2.6 (1.8)	2.4 (2.0)	304	UK	All participants also had supervised daily toothbrushing.	Strong evidence of differences in BI at the 3, 6- and 12-month time-points ($p < 0.01$) were not observed at 18 or 24 months

Footnotes

Shaw 1991; Data also reported for 3, 6, 12 and 18 months, not reported here.

Indices: WHOB I; World Health Organisation Bleeding Index, Scale 0 -1; presence or absence of bleeding after 20 seconds.

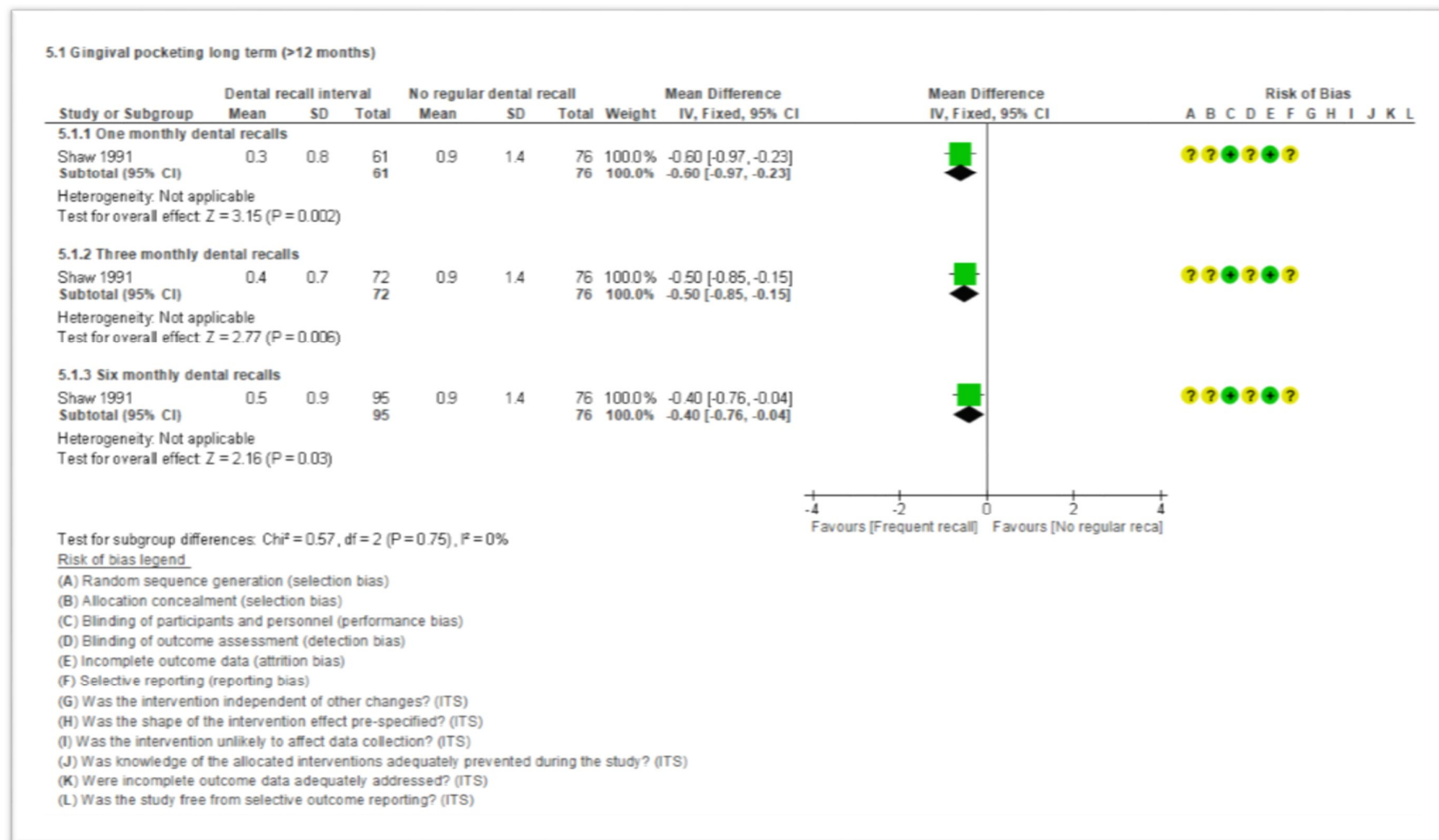


Figure 27: C5 Meta-analysis Gingival inflammation – long term

5.2 Gingival bleeding long term (>12 months)

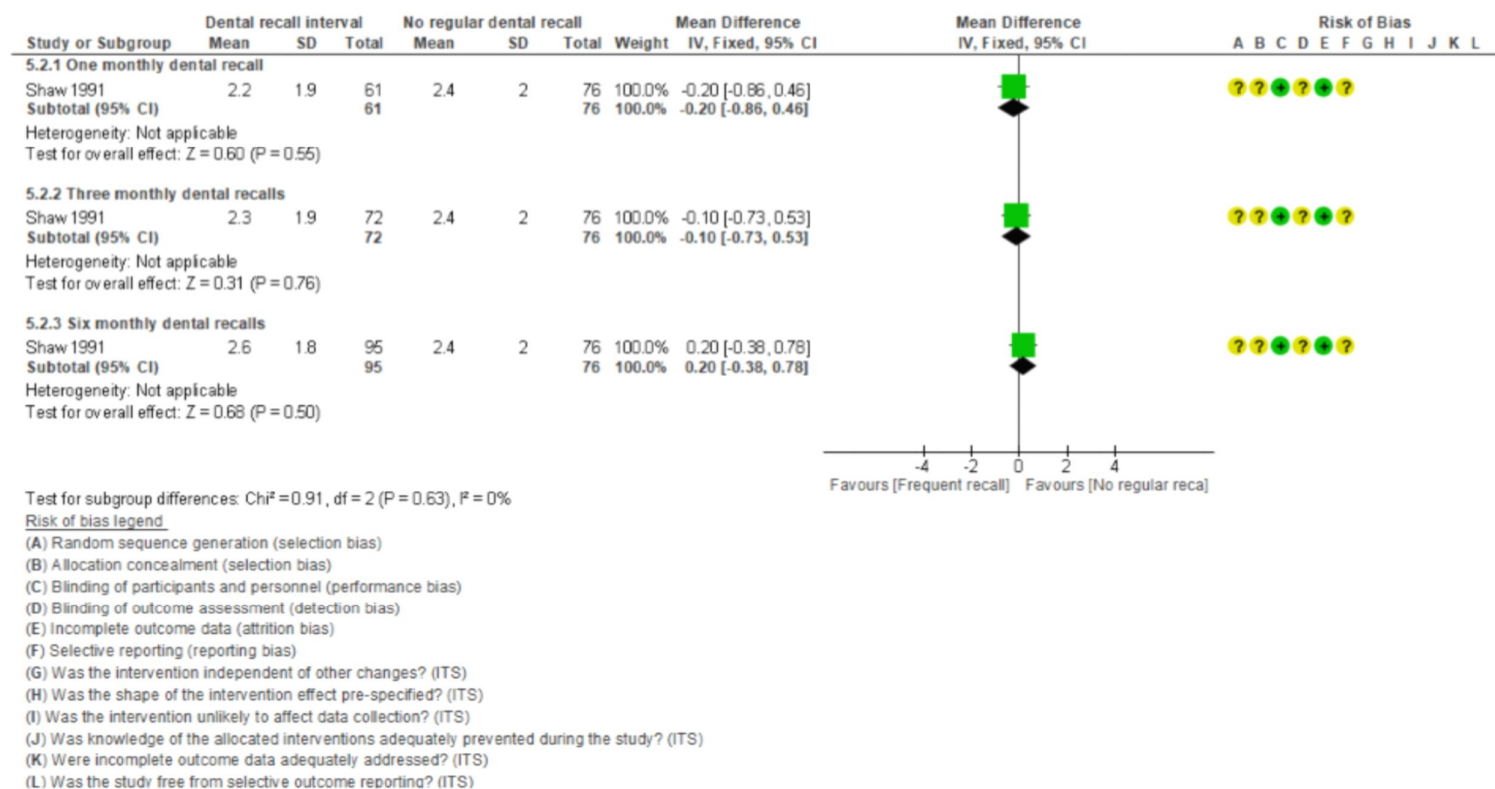


Figure 28: C5 Meta-analysis Gingival bleeding – long term

Table 50: C5 Plaque – long term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
24 months	Shaw 1991	One, three- and six-monthly dental recall versus no regular dental recall	WHOPI (Scale 0-3)	R1 mth: 1.2 (1.7) R3 mth: 1.3 (1.7) R6 mth: 1.3 (1.5)	2.0 (1.9)	304	UK	All participants also had supervised daily toothbrushing.	Strong evidence of differences for PI at 12 (p < 0.01) and 24 months (p < 0.05), the greatest differences were seen in the 1 and 3 monthly intervals.

Footnotes

Shaw 1991; Data also reported for 3, 6,12 and 18 months, not reported here. " the difference between the one-monthly and three-monthly hygienist input (Recall visit) were not clinically very apparent after two years." Pg. 144

Indices: WHOPI; World Health Organisation Plaque Index; data are for "Mean number of sextants scored as Code 2"

Table 51: C5 Calculus – long term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
24 months	Shaw 1991	One, three- and six-monthly dental recall versus no regular dental recall	WHOCI (Scale 0 -1)	R1 mth: 1.6 (1.7), R3 mth 2.2 (1.9), R6 mth: 3.5 (2.0)	3.6 (2.1)	304	UK	All participants also had supervised daily toothbrushing.	Strong evidence of differences in CI at 6, 12, 18 and 24 months (p < 0.01).

Footnotes

Shaw 1991; Data also reported at 3, 6,12 and 18 months, not reported here.

Indices: WHOCI; World Health Organisation Calculus Index, Scale 0 -1 (presence or absence).

5.3 Plaque long term (> 12 months)

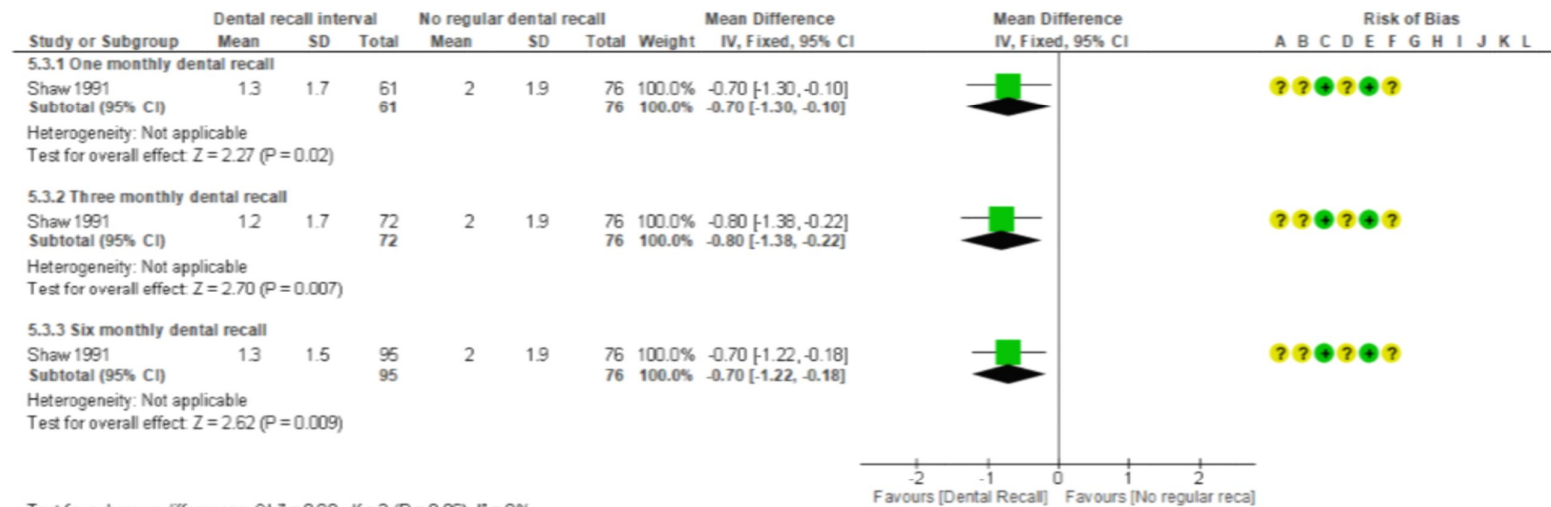
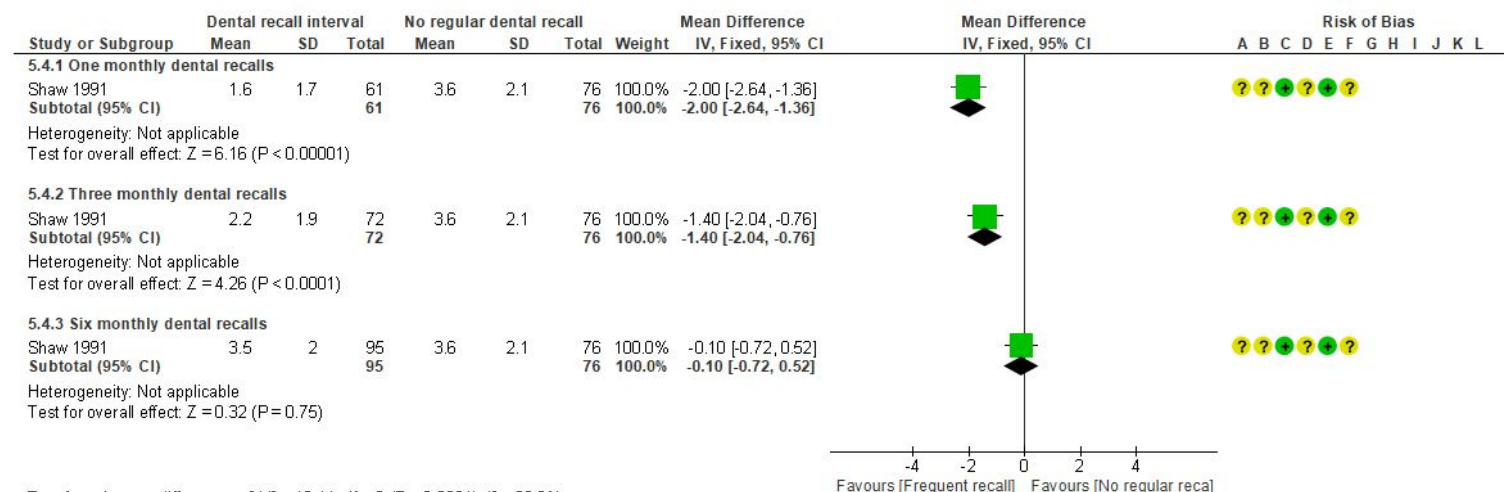


Figure 29: C5 Meta-analysis Plaque – long term

5.4 Calculus long term (> 12 months)



Risk of bias legend

- (A) Random sequence generation (selection bias)
- (B) Allocation concealment (selection bias)
- (C) Blinding of participants and personnel (performance bias)
- (D) Blinding of outcome assessment (detection bias)
- (E) Incomplete outcome data (attrition bias)
- (F) Selective reporting (reporting bias)
- (G) Was the intervention independent of other changes? (ITS)
- (H) Was the shape of the intervention effect pre-specified? (ITS)
- (I) Was the intervention unlikely to affect data collection? (ITS)
- (J) Was knowledge of the allocated interventions adequately prevented during the study? (ITS)
- (K) Were incomplete outcome data adequately addressed? (ITS)
- (L) Was the study free from selective outcome reporting? (ITS)

Figure 30: C5 Meta-analysis Calculus – long term

Table 52: C5 All outcomes and subgroup effect estimates

5 Dental recall intervals

Outcome or Subgroup	Studies	Participants	Statistical Method	Effect Estimate
5.1 Gingival pocketing long term (> 12 months)	1		Mean Difference (IV, Fixed, 95% CI)	Subtotals only
5.1.1 One monthly dental recalls	1	137	Mean Difference (IV, Fixed, 95% CI)	-0.60 [-0.97, -0.23]
5.1.2 Three monthly dental recalls	1	148	Mean Difference (IV, Fixed, 95% CI)	-0.50 [-0.85, -0.15]
5.1.3 Six monthly dental recalls	1	171	Mean Difference (IV, Fixed, 95% CI)	-0.40 [-0.76, -0.04]
5.2 Gingival bleeding long term (> 12 months)	1		Mean Difference (IV, Fixed, 95% CI)	Subtotals only
5.2.1 One monthly dental recall	1	137	Mean Difference (IV, Fixed, 95% CI)	-0.20 [-0.86, 0.46]
5.2.2 Three monthly dental recalls	1	148	Mean Difference (IV, Fixed, 95% CI)	-0.10 [-0.73, 0.53]
5.2.3 Six monthly dental recalls	1	171	Mean Difference (IV, Fixed, 95% CI)	0.20 [-0.38, 0.78]
5.3 Plaque long term (> 12 months)	1		Mean Difference (IV, Fixed, 95% CI)	Subtotals only
5.3.1 One monthly dental recall	1	137	Mean Difference (IV, Fixed, 95% CI)	-0.70 [-1.30, -0.10]
5.3.2 Three monthly dental recall	1	148	Mean Difference (IV, Fixed, 95% CI)	-0.80 [-1.38, -0.22]
5.3.3 Six monthly dental recall	1	171	Mean Difference (IV, Fixed, 95% CI)	-0.70 [-1.22, -0.18]
5.4 Calculus long term (> 12 months)	1		Mean Difference (IV, Fixed, 95% CI)	Subtotals only
5.4.1 One monthly dental recalls	1	137	Mean Difference (IV, Fixed, 95% CI)	-2.00 [-2.64, -1.36]
5.4.2 Three monthly dental recalls	1	148	Mean Difference (IV, Fixed, 95% CI)	-1.40 [-2.04, -0.76]
5.4.3 Six monthly dental recalls	1	171	Mean Difference (IV, Fixed, 95% CI)	-0.10 [-0.72, 0.52]

Comparison 6: Use of individual clinical photographs as a toothbrushing motivator versus no motivator for people with ID

6.1 Plaque (medium term)

One RCT, with 29 participants, compared the use of discussing clinical photographs of their disclosed dental plaque with the participants, taken at monthly oral hygiene sessions during the intervention, with no discussion of clinical photographs, on plaque levels over a period of six months (Bickley 1990), (Table 53).

There was no difference found in plaque levels between the intervention and control (MD: -0.10, 95% CI: -1.85 to 1.65); there was a small difference in the extrinsic stain score in favour of the intervention (MD: -2.60, 95% CI: -19.45, 14.25), (Figure 31). The certainty of the evidence is low. The effect estimates for all outcomes and subgroups included in the Forest Plots for Comparison 6 are presented in Table 54.

Comparison 7: Daily, twice weekly and once weekly brushing frequency versus usual care for people with ID

Gingival inflammation (short term)

One RCT, with 80 participants, compared the frequency of brushing over a period of 21 days, on gingival inflammation (Swallow 1969), (Table 55). Daily brushing was compared to twice weekly, once weekly or usual care. This study also compared the use of an electric toothbrush and a manual toothbrush (reported earlier).

For people with ID, who have their teeth brushed by a dental professional, the more frequently the teeth are brushed the greater the reduction in gingival inflammation in the short term (21 days). The certainty of the evidence is moderate.

Table 53: C6 Plaque – medium term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
6 months	Bickley 1990	Discussion of clinical photographic records versus no discussion	WHOPI (Scale 0 - 12)	7.3 (2.8)	7.4 (2.0)	29	UK	Disclosing solution was also used	"There were no significant changes after 6 months in either of the groups." Pg. 4
6 months	Bickley 1990	Discussion of clinical photographic records versus no discussion	ESI (%)	37.7% (18.2)	40.3% (26.4)	29	UK	Disclosing solution was also used	"the test group was considerably lower but just failed to achieve statistical significance (t = 1.721, df =15)" Pg. 4

Footnotes

Indices: WHOPI; World Health Organisation Plaque Index (4 surfaces per tooth, scored 0 - 3 resulting in a Scale of 0 - 12). ESI; Extrinsic Stain Index Shaw & Murray (% Plaque on anterior teeth).

Table 54: C6 All outcomes and subgroup effect estimates

6 Clinical photographs as OH motivators				
Outcome or Subgroup	Studies	Participants	Statistical Method	Effect Estimate
6.1 Plaque medium term (6 weeks to 12 months)	1		Mean Difference (IV, Fixed, 95% CI)	Subtotals only
6.1.1 Plaque	1	29	Mean Difference (IV, Fixed, 95% CI)	-0.10 [-1.85, 1.65]
6.1.2 Extrinsic stain	1	29	Mean Difference (IV, Fixed, 95% CI)	-2.60 [-19.45, 14.25]

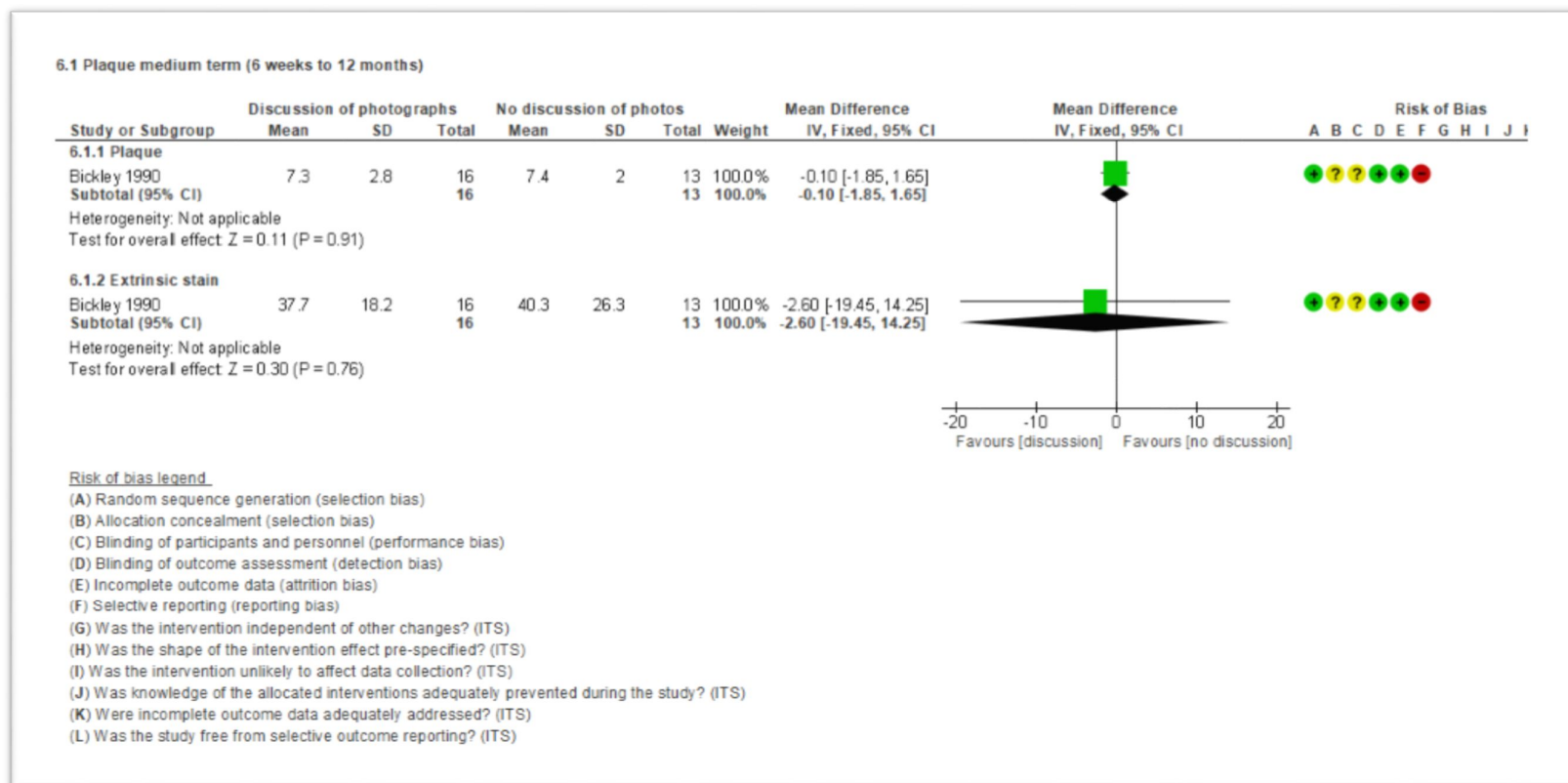


Figure 31: C6 Meta-analysis Plaque and Extrinsic stain – medium term

Table 55: C7 Gingival inflammation – short term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
21 days	Swallow 1969	Daily V Twice Weekly brushing	B&LL&SGI (Scale 0 - 6)	Pooled 1.84 , Electric 1.24, Manual 2.43	Pooled 2.05 , Electric 1.59, Manual 2.51	40	UK	Pooled mean calculated from reported data.	Brushing twice a week reduced gingival disease by approx. 46% and brushing daily by 51%. Pg. 378
21 days	Swallow 1969	Daily V Once Weekly brushing	B&LL&SGI (Scale 0 - 6)	Pooled 1.84 Electric 1.24, Manual 2.43	Pooled 3.42 Electric 3.26 Manual 3.57	40	UK	Pooled mean calculated from reported data.	Brushing one a week reduced gingival disease by approx. 26% and brushing daily by 51%. Pg. 378
21 days	Swallow 1969	Daily brushing V Usual Care	B&LL&SGI (Scale 0 - 6)	Pooled 1.84 Electric 1.24, Manual 2.43	4.27	60	UK	Pooled mean calculated from reported data.	Brushing daily differed strongly from Usual Care - decreasing levels of gingivitis by 51%. Pg. 378

Footnotes

Swallow 1969; Data were presented separately for electric and manual toothbrushes, labial and buccal surfaces and males and female, no SD reported. The data for the females using the electric toothbrush daily and twice weekly appears unrealistically low, author could not be contacted to clarify. Pooled means were calculated from these data. Usual Care - "rarely received any regular form of oral hygiene".

Indices: B&LL&SGI; Buccal and Labial surfaces Loe & Silness: Scale 0 - 6.

Comparison 8: Use of a toothpaste with a plaque disclosing agent versus a conventional toothpaste for people with ID

Gingival Inflammation and Plaque (short term)

One non-randomised, two-armed cross-over trial, with 40 participants, compared the use of a toothpaste containing a plaque disclosing agent with a conventional toothpaste, on gingival inflammation and plaque levels over a period of ten days (Teitelbaum 2009).

For people with intellectual disabilities, the use of a toothpaste containing a plaque disclosing agent when brushing, may reduce plaque and gingival inflammation in the short term (ten days), (Table 56; Table 57). The certainty of the evidence is very low.

Comparison 9: Individualised oral care plan versus usual care for people with ID

Plaque (medium term)

One NRCT, with 79 participants, compared the use of an individualised oral care plan with usual care, on plaque levels, over a period of 12 months (Altabet 2003), (Table 58). Based on the findings of this study, an individualised oral care plan may reduce plaque levels for people with intellectual disabilities over a period of 12 months. The certainty of the evidence is low.

Table 56: C8 Gingival inflammation – short term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
10 days	Teitelbaum 2009	TP with a disclosing agent versus a regular TP	GBI (Scale 0 - 1)	% Mean Diff 18%	% Mean Diff 8%	40	Brazil	Cross-over trial. Data are presented for two arms combined.	There was strong evidence of a difference in the bleeding index in favour of the intervention (p < 0.001) Pg. 466

Footnotes

Teitelbaum 2009; Included two other experimental groups using Chlorhexidine, not relevant to this review. The toothpaste containing a plaque disclosing agent produced a reduction in gingival inflammation similar to the dentifrice with chlorhexidine (18%) (not reported here). Exact data in Figure 2 unclear, requested from authors, no response.

Indices: GBI; Gingival Bleeding Index Ainamo & Bay (present or absent).

Table 57: C8 Plaque – short term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
10 days	Teitelbaum 2009	TP with a disclosing agent versus a regular TP	SG&VOHI (Scale 0 - 3)	% Mean Diff 65%	% Mean Diff 15%	40	Brazil	Cross-over trial. Data are presented for two arms combined.	There was strong evidence of a difference in plaque levels in favour of the intervention. (p < 0.0001) Pg. 466

Footnotes

Teitelbaum 2009; Included two other experimental groups not relevant to this review. Exact data in Figure 2 unclear. Requested from authors, no response.

Indices: SG&VOHI - Simplified Green & Vermillion Oral Hygiene Index - plaque only. Scoring of disclosed plaque on buccal surfaces of 4 teeth and lingual surfaces of 2 teeth.

Table 58: C9 Plaque – medium term

Duration	Study ID	Comparison	Index	Intervention Mean (SD)	Control Mean (SD)	Total No.	Country	Comments	Summary
12 months	Altabet 2003	Individualised oral care plan versus no care plan.	SOHR (Scale 1-5)	2.97	3.10	79	USA	The intervention group was monitored, monthly - quarterly, with staff feedback and review as required. No monitoring of the control group.	"People in the treatment group showed statistically significant improvement, t (39) = 3.82, p < .001)" Pg. 442
12 months	Altabet 2003	Individualised oral care plan versus no care plan.	Improvement differential	38%	5%	79	USA	The intervention group was monitored, monthly - quarterly, with staff feedback and review as required. No monitoring of the control group.	"The results indicate that practices can be implemented to improve oral hygiene in individuals with mental retardation living at a state residential facility." Pg. 442

Footnotes

Indices: SOHR; Subjective Oral Hygiene Rating; Scale 1 - 5 (1 excellent: no plaque on teeth, 2 good; plaque present on less than 15% of total tooth surface (tts), 3 fair; plaque present on 15 - 50% of tts, 4 poor; plaque present on 50 - 90% of tts, 5 plaque present on greater than 90% of tts.) Pg. 441. Numerical ratings were averaged over the two designated time periods. Improvement Differential; difference between percentage of individuals with improvement rating minus the % individuals with poorer rating over same time period.

4.8 Summary of main results

This review reported findings for nine different types of oral hygiene interventions targeting people with intellectual disabilities or their carers. Four of the intervention types reviewed between six and thirteen studies each. The remaining five intervention types reviewed only one study each. The individual studies were generally small: many studies having less than 50 participants, which resulted in most of the meta-analyses having less than 100 participants in total. The main outcomes reported were gingival health, oral hygiene knowledge and behaviour in relation to oral hygiene routines of both people with ID and their carers. No studies reported on dental caries or quality of life issues.

Comparison 1. Special manual toothbrushes versus conventional manual toothbrushes (6 RCTs and 2 NRS)

There was low quality evidence in one RCT medium term (< 12 months) in relation to gingival inflammation, when comparing the use of a special manual toothbrush (Superbrush) to a conventional manual toothbrush, indicating a small effect size in favour of a special manual toothbrush at reducing levels of gingival inflammation (Table 18,), (Figure 7) and improving behaviour (Table 23), (Figure 11, Figure 12). In this study the toothbrushing was carried out by the carers. There was no difference in effect size between the two different toothbrushes for all the other outcomes and time frames measured.

One ITS study compared the Collis Curve toothbrush to a conventional toothbrush, medium term, and found no difference between the two toothbrushes (Table 21). No studies reported on dental caries.

Comparison 2. Electric toothbrushes versus manual toothbrushes (8 RCTs and 5 NRS)

There was moderate quality evidence in two RCTs in relation to the assessment of gingival inflammation, plaque and calculus in the medium term when comparing electric and manual toothbrushes, indicating that there was little or

no difference between the two different types of toothbrushes (Table 26, Table 30, Table 33), (Figure 13, Figure 15, Figure 17).

The findings in the NRS were broadly similar to the findings of the meta-analysis but the certainty of the evidence was very low (Table 27, Table 28, Table 31, Table 32), (Figure 14, Figure 16). The plaque and gingival inflammation levels of the control group in all these NRS studies improved from baseline, which was credited to the daily brushing routine instigated as part of the studies.

The evidence in relation to gingival inflammation and plaque, short term, was inconclusive as the findings were inconsistent between the four studies; the data could not be included in a meta-analysis (Table 25, Table 29).

Dental caries and quality of life were not reported in any of the studies. The range of electric toothbrushes, with a variety of modes of action, was diverse. The difference in findings between the types of electric toothbrush used may be relevant.

Comparison 3. Oral hygiene care training of carers versus no oral hygiene care training of carers (4 RCTs and 3 NRS)

Oral hygiene care training for carers resulted in a small reduction in the level of gingival inflammation of the people with ID for whom they cared, in the RCTs medium term, compared to no oral hygiene care training, the quality of the evidence was low (Table 35), (Figure 18).

The plaque levels of the people with ID for whom the carers cared, showed no evidence of a difference between intervention and control groups (Table 36), (Figure 19), the quality of the evidence was low. One NRS did show strong evidence of a difference in plaque levels short term, in favour of the intervention, when the carers were made accountable for the level of care they provided (Table 37), (Figure 20), the quality of the evidence was very low.

Changes in carers knowledge generally showed a moderate improvement following training of carers (Table 39), (Figure 21, Figure 22). There was a

small positive change in attitude, self-efficacy and behaviour, however, the findings between studies was inconsistent (Table 40; Table 41), (Figure 23). The quality of evidence in the RCTs was low for these outcomes. Issues such as level of attendance at training, use of the educational resources, instruction on how to apply the training in practice and levels of support provided to carers were identified as possibly impacting on the outcomes.

Comparison 4. Oral hygiene training of people with ID versus no training of people with ID (2 RCTs and 4 NRS)

Small non-significant and moderate significant reductions in the levels of gingival inflammation and plaque levels, respectively, in favour of oral hygiene training of people with ID compared to no oral hygiene training, were observed (Table 44; Table 45), (Figure 24, Figure 25). The quality of the evidence was low.

Change, in favour of the intervention, was also seen in three of the four NRS assessing changes in behaviour, as measured by an increase in the steps achieved in a toothbrushing routine. The quality of the evidence was very low (Table 46), (Figure 26). It is unclear if the improvement in the tooth brushing routines in the NRS impacted on gingival health, as no clinical measurements were carried out.

Comparison 5. Dental recall intervals versus usual care for people with ID (1 RCT)

One RCT had low quality evidence that more regular dental recall visits might improve gingival health (gingival pocketing, gingival bleeding, plaque and calculus levels) in the medium and long-term (Table 48; Table 49; Table 50; Table 51), (Figure 27, Figure 28, Figure 29, Figure 30). The difference between one, three- and six-monthly recall intervals over a 24-month time frame was minimal and unlikely to be clinically significant.

Comparison 6. Use of clinical photographs as an oral hygiene motivator for people with ID versus no motivator (1 RCT)

One RCT had low quality evidence demonstrating that the use of clinical photographs as an oral hygiene motivator most likely had no effect on plaque levels in the short term (Table 53), (Figure 31).

Comparison 7. Daily, twice weekly and one weekly tooth brushing versus usual care for people with ID (1 RCT)

One RCT had moderate quality evidence demonstrating that brushing daily compared to brushing twice weekly, once weekly or usual care may reduce plaque levels in the short term, the brushing was carried out by a dental professional in this intervention (Table 55). Meta-analysis of the data was not possible.

Comparison 8. Use of a toothpaste with a plaque disclosing agent versus a conventional toothpaste (1 NRS)

One NRS found that the use of a toothpaste with a plaque disclosing agent might reduce plaque and gingival inflammation levels in the short term (Table 56, Table 57). The quality of the evidence was very low.

Comparison 9. Individualised oral care plans versus usual care (1 NRS)

One NRS found that individualised oral care plans might improve oral hygiene in the medium term (Table 58). The intervention involved the input of a multi-disciplinary team. The quality of the evidence was very low.

4.9 Discussion

Oral hygiene interventions, specifically, are one of the most common types of oral health interventions targeting those with disabilities (Waldron *et al* 2016). The clinical and behavioural outcomes assessed in these interventions included: the levels of gingival inflammation, plaque and calculus of the people with ID and the levels of knowledge, behaviour, attitude and self-efficacy of both the people with ID and their carers. The aim of this review was to determine the effects (benefits and harms) of oral hygiene interventions for people with ID.

Following an extensive search of the literature, 34 studies were included in this Cochrane Review (19 RCTs and 15 NRS), which were reported between 1963 and 2016. The studies included 1795 participants with ID, most of the studies included participants with mixed levels of ID. The participants were mostly adults with ages ranging from 18 to 64 years. Only seven studies targeted the carers of the people with ID (354 participants). The individual studies were generally small: many studies having less than 50 participants, which resulted in most of the meta-analyses having less than 100 participants in total.

Thirteen studies compared electric toothbrushes to manual toothbrushes, eight studies compared a special manual toothbrush with a conventional toothbrush, seven studies focused on the training of carers to improve the oral health of the people with ID for whom they cared, six studies focused on the training of people with ID in the practical skills of toothbrushing to improve their oral health, the five remaining studies each had a unique intervention.

The length of follow-up of the interventions overall was relatively short, only three studies were followed-up for more than 12 months, twenty studies had a follow-up period of more than 6 weeks, but less than 12 months and ten studies were followed-up for 6 weeks or less. The duration of one study was unclear.

4.9.1 Overall completeness and applicability of evidence

The primary outcome of gingival health was reported using a variety of indices measuring gingival inflammation (18 studies), plaque (20 studies) and calculus (4 studies). The indices used to assess these outcomes were modified or adapted for the population. Any persistent resistance to the clinical assessments or to the intervention, by the people with ID, resulted in them being excluded from the study. Any differences observed in the clinical assessments must be considered in relation to their practical impact on gingival health long term. Reporting the effect size as well as the statistical significance of the findings allows the magnitude or size of the difference to be transparent, allowing the reader to quantify the practical significance of a finding. The size of the sample, the importance of the finding and confidence interval must also be considered.

Of the clinical secondary outcomes, no studies provided data on dental caries, although one study reported assessing it. The non-clinical secondary outcomes reported for the carers were knowledge, behaviour, attitude and self-efficacy. Only changes in behaviour were reported for the people with ID.

One study reported on carers' experiences when brushing the teeth of the people with ID for whom they cared, when using two different types of toothbrushes and also the level of resistance to the toothbrushing, by the people with ID who were having their teeth brushed.

The clinical and behavioural outcomes focused predominately on the benefits. Any harms identified were not quantified, tended to be reported in the discussion section, were transient and not considered as a formal outcome. Any formal qualitative assessments of the interventions were completed by the carers involved and were normally in the form of post study interviews.

Quality of life changes and long term, patient-centered outcomes were not addressed formally by any of the studies identified in this review. There were occasional comments in the discussion section of the studies regarding increases in self-efficacy or confidence of the carers. The lack of qualitative outcomes may be linked to the age of many of the studies and lack of reporting standards for the earlier studies in this review.

In relation to unintended effects, no studies reported any positive unintended effects. Negative unintended effects were recorded in the characteristics of studies tables under the heading of adverse effects: the few studies that reported adverse effects only reported them within the text, no formal assessments were reported. The most common adverse effects reported related to difficulties encountered when using different types of toothbrushes.

The topics considered for subgroup analysis, as outlined in the protocol, were recorded for the individual studies. The only subgroup topic with sufficient data to make a subgroup comparison was the studies duration (short, medium and long-term). There were insufficient data to make any other useful analyses.

4.9.2 Validity of assessments used

All the clinical outcomes assessed used validated tools, except for six studies that used subjective assessments (Albino 1979; Gertenrich 1967a; Gertenrich 1967b; Gertenrich 1967c; Gertenrich 1967d; Kelner 1963). Many of the validated tools were, however, modified, some to a considerable extent; some of these modifications have been validated and some have not. The level of modifications to the assessment tools highlights the difficulties involved in carrying out clinical assessments for this population.

The use of subjective clinical assessments is most likely an historical issue and unlikely to occur in any future interventions. Despite the subjective nature of these assessments, these studies are considered to add value to this review; their findings have been graded with the quality of these assessments in mind.

The non-clinical outcomes; knowledge, behaviour, attitude and self-efficacy, were less well validated. The studies that included observations of behaviour were assessed for reliability. One study, using a questionnaire to assess knowledge, self-efficacy, attitude and behaviour, reported that the tool had been validated; another study used the same tool. Another study, using a questionnaire, reported getting "expert advice on the content".

4.9.3 COM-B Characteristics identified in the interventions

The behavioural change elements of the interventions in this review, were mapped using the COM-B framework for understanding behaviour and behaviour change (Michie *et al* 2011), as these elements are regarded as a reliable way of predicting behavioural change (Newton & Asimakopoulou 2015). These elements are: Capability, defined as the psychological and physical capacity to engage in the activity. Opportunity, both physical and social, which considers contexts beyond the individual and Motivation, both automatic and reflexive, which might result in conscious decision-making. An intervention may change one or more of these elements resulting in the desired behaviour of, for example, performing regular oral hygiene.

Capability

The potential for increasing the physical capability of the participants, was identified in interventions that focused on training, modelling or enablement in the skill of toothbrushing.

The potential for increasing the psychological capability was predominantly identified in interventions for people with mild or moderate ID as they could have the capability to reflect and to make the connections between any information provided and the benefit to them. Interventions targeting people with more severe levels of ID may benefit more from interventions that use incentivisation, persuasion, coercion or restriction as a psychological tool.

Opportunity

The element of opportunity to change behaviour was considered, for example, if the intervention attempted to provide additional time or resources to the oral hygiene routine (physical opportunity) or to make the oral hygiene routine socially expected or the cultural norm (social opportunity).

Physical opportunity most often involved specific time being set aside for the toothbrushing routine, environmental restructuring such as providing different oral hygiene or other aids and changing the setting where the toothbrushing took place.

The social opportunities identified were monitoring or recording of the oral hygiene routine: such as a toothbrushing diary or staff monitoring. Social opportunity-based interventions tended to focus on carers rather than people with ID. One example of an intervention that did focus on children with ID, involved a toothbrushing session using a plaque disclosing solution in a classroom setting; the children were aware of how their classmates were performing, which may have acted as a social cue in relation to the toothbrushing habit (Shaw *et al* 1983).

Motivation

The element of motivation to change behaviour was considered if the intervention included the potential to reflect on the benefit of changing

behaviour or for the behavioural change to become an automatic process. It was the least common element identified in the interventions.

Reflective motivation was identified in interventions that included some element of feedback in relation to behaviour or performance and was seen predominantly in the interventions targeting carers.

No studies were identified as having the potential for the behavioural change to become automatic. This may have been due to the duration of the interventions, as the development of habit can take some time; 19 of the studies were followed up for three months or less and only three studies for more than 12 months.

To ensure that a successful intervention is replicated in a new setting, the explicit causal processes that elicited the successful behavioural change must be properly identified and replicated. It is difficult to identify the COM-B elements retrospectively, we can only claim that the intervention may have provided these elements. The decision to retrospectively identify these elements in our review provided valuable insight into the causal processes that were potentially in action. By mapping the behavioural change elements and intervention functions inherent in the interventions in this review, we hope we have assisted in the identification of the causal processes that should be replicated. These details are recorded in the individual Characteristics of included studies tables (Appendix 4).

4.9.4 Quality of the evidence

The risk of bias for the RCTs and NRCTs was assessed using the standardised risk of bias criteria (Higgins 2011a). The ITS and RM studies were assessed for risk of bias using the EPOC seven standard criteria (EPOC 2016). The certainty of the evidence was determined using the GradePro software under the following headings: study design (risk of bias); inconsistency; indirectness; imprecision; publication bias; large effect; plausible confounding; dose response gradient (Schünemann 2013). The quality of the body of evidence for

each of the main outcomes for each comparison was categorised as high, moderate, low or very low.

The certainty of the evidence varied from moderate to very low for comparisons with RCT evidence and was very low for comparisons assessed only in NRS. The quality of reporting was poor; the lack of details provided in relation to the methodology impacted on the risk of bias. More than half of the studies were pre-2000 and thus undertaken before the introduction of guidelines such as the CONSORT statement in relation to the standards for reporting trials (Moher 2001, Schulz *et al* 2010). The inclusion of NRS, which was considered important due to the likely use of these study designs because of the complexity of the interventions, also reduced the quality of the evidence.

4.9.5 Potential biases in the review process

The most common element reducing the quality of the evidence was the 'unclear' elements under all the risk of bias headings, most notably in relation to outcome reporting, which was predominantly as a consequence of the absence of study protocols. The level of 'high' risk in relation to randomisation was caused by the inclusion of non-randomised studies. The decision to include these non-randomised studies was based on concerns regarding the complexities of interventions targeting people with ID and is discussed later in this chapter.

Sixteen studies were excluded as the articles could not be sourced, we were unable to contact the authors for more details or, if they were contactable, the data were no longer available. These studies, if eligible for inclusion, may have changed the findings of the Review, however, as the type of interventions addressed by them were spread over the entire range of intervention types reviewed, their impact on the conclusions would most likely have been minimal.

As the first review of this topic, we have worked hard to identify all relevant studies but acknowledge that some studies may not have been published,

registered or presented anywhere and we did not search every database in existence.

The study designs included in this review are wider than conventional Cochrane Reviews, but have allowed the consideration of interventions that might otherwise have been excluded. It was suspected, at the protocol stage, that the diverse nature of ID, the related co-morbidities, the environments and level of supports available that combine to create a complex population, would favour before-and-after study designs. However, this assumption was not borne out, as a small number of RCTs were identified for most of the interventions.

Given the number of studies reporting similar or no change for the intervention group compared to the controls, it is unlikely that there is a publication bias for positive findings in the studies being reported.

4.9.6 Agreements and disagreements with other studies or reviews

The findings of our review can be compared to a variety of other Cochrane Oral Health reviews, other systematic reviews or relevant studies targeting the general population.

Special manual toothbrushes

The comparison of special manual toothbrushes to conventional manual toothbrushes in our review can be compared to the systematic review by Kalf-Scholte (2018), of triple-headed toothbrushes compared to single-headed toothbrushes, used in a general population ("humans without fixed orthodontic appliances"), in relation to the outcome of prevalence of plaque, which included three of the studies included in our review (Dôgan 2004; Kaschke 2005; Sauvetre 1995). Both our review and that of Kalf-Scholte (2018), noted some subgroup analysis which demonstrated that if the toothbrushing is assisted or carried out by a carer, there may be a greater benefit to using a triple-headed toothbrush ('Superbrush').

A meta-analysis carried out in our review included just one study, which was not included in the Kalf-Scholte 2018 review (Bildt 2010). In the Bildt study the toothbrushing was carried out by the carers and also showed a small effect size in favour of the special manual toothbrush. The Kalf-Scholte 2018 review concluded that, overall, both brushes are equally effective for self-brushers; a finding that matches the overall findings of our review.

Electric toothbrushes

The findings of the comparison between electric toothbrushes and manual toothbrushes in our review appear to conflict with the data from the Yaacob (2014) review in a general population, reviewing the use of powered versus manual toothbrushes. The Yaacob (2014) review concluded that electric toothbrushes were more effective than manual toothbrushes both short (1 to 3 months) and long (> 3 months) term. The review reported a moderate effect size in favour of the electric toothbrush, short term (SMD -0.50, 95% CI -0.70 to -0.31) and long term (SMD -0.47, 95% CI -0.82 to -0.11). Our review showed no evidence of a difference for gingival inflammation levels between toothbrushes at six months (MD: 0.02, 95% CI: -0.06 to 0.09) (Figure 13). This may be due to differences in the toothbrushing skills of the participants (people with ID or their carers), regardless of the type of toothbrushing being used, or the type of electric toothbrush being used, or may be due to adverse effects encountered by the participants in the studies.

The greatest differences in the Yaacob (2014) review were seen in the studies that compared an electric toothbrush with a rotating oscillation mode of action. There were only two studies in our review that compared an electric toothbrush with a rotation oscillation mode of action (Braun Plaque Control 3D) to a manual toothbrush (Bratel 1991; Dôgan 2004). The RCT (Dôgan 2004) found that there was strong evidence of a difference in plaque levels in favour of the electric toothbrush after one week ($P < 0.001$). The interrupted time series study (ITS), (Bratel 1991) found some strong evidence of differences in sub groups for plaque at 12 weeks (those using the electric toothbrush unaided) and gingivitis at 16 months (those using the electric toothbrush unaided) in favour of the electric toothbrush ($P < 0.05$). The data for these studies were not suitable for meta-analyses.

Oral hygiene care training of carers

Comparisons, in relation to changes in carers' knowledge, attitude, self-efficacy and behaviour in our review, can be made to studies directed at carers in long-term healthcare settings. A systematic review of the effectiveness of continuing education in long term care settings by Aylward *et al* (2003) found many of the same issues in their review in relation to the quality of the evidence, diversity of assessment tools, level of attrition, applying the training in practice and levels of support provided to carers at management level. Of the studies in their review that assessed both knowledge and behaviour, almost all showed an improvement in knowledge but no change in behaviour. Most of the evaluations took place immediately after the intervention, which ranged in length from 10 mins to 52 hours.

The clinical outcomes assessed in this comparison in our review, can be compared to the Cochrane Review by Albrecht *et al* (2016), which reviewed oral health education interventions by nurses in nursing homes. These authors concluded that there was no difference in plaque levels after such interventions compared to usual care. The two RCTs in our review similarly found no difference in plaque or gingivitis levels. Two NRS in our review did show some reduction in plaque levels when staff were made accountable for the oral hygiene care of people with ID.

Oral hygiene care training for people with ID

The RCTs included in our review comparing oral hygiene care training for people with ID were all short-term studies. The findings are in agreement with the systematic review by Watt *et al* (2005) in relation to oral health promotion in general, which concluded that short-term interventions may results in a reduction in plaque and gingivitis levels. However, they noted in their review that there was very limited evidence supporting long-term reductions in plaque and gingival bleeding indices as a consequence of training. The NRS in our review, which were medium- and long-term interventions did show some improvement in toothbrushing routines. However, these studies had small numbers of participants and were very labour intensive. Kay & Locker (1996)

commented in their review, in relation to oral health promotion, that the balance between input and output must be considered.

Recall intervals

The findings of the one intervention we cited comparing recall intervals and their impact on oral health outcomes may be compared to the published Cochrane Review on routine scale and polish for periodontal health in adults (Worthington *et al* 2013, Lamont *et al* 2018). Although, the participants levels of gingival inflammation in the study in our review was slightly higher than that of the participants in the two studies included in the review of the general population. Our review found that the more regular the recall interval (1, 3 or 6-monthly) the greater the benefit to gingival health. However, the certainty of the evidence was low. Worthington and colleagues (2013, 2018) found little or no evidence to support more regular scale and polish visits (6 monthly), and also found the number of studies and the quality of the evidence poor.

4.10 Conclusion

This review found evidence that a special manual toothbrush (the 'Superbrush') may have a greater effect on the reduction of levels of gingival inflammation in the medium term, when compared to a conventional manual toothbrush, for people with ID, when the toothbrushing was carried out by carers. A special manual toothbrush (the 'Superbrush') may also improve the quality of life of people with ID and their carers; the brush was found to be easier to use by carers, resulting in a more positive experience for them, there was less resistance by people with ID to having their teeth brushed and there was an increase in the frequency of brushing episodes.

The review also showed that training carers in oral hygiene care for people with ID may increase carers oral hygiene knowledge and may improve their behaviour, attitude and self-efficacy in relation to providing oral hygiene care for the people for whom they care. This carer training may also reduce the levels of gingival inflammation of the people with ID for whom they care.

Additionally, oral hygiene training for people with ID, may reduce their plaque levels, short term.

The benefits of using an electric toothbrush compared to a manual toothbrush for people with ID was inconclusive. There were also insufficient studies to compare the different types/modes of action of electric toothbrushes.

The limitations of this review must be acknowledged. Overall, the quality of the studies identified in this review and the manner in which the evidence was reported, was poor. This prevented the inclusion of many of the studies in meta-analyses, thus reducing the certainty of the evidence.

In particular, the data from several cross-over studies, which should have been a useful source of data, were excluded from the meta-analyses, due to the type of data reported. For studies using cross-over trials, the mean difference or data from each treatment period should be reported to allow calculation of the standard error of the mean difference or to approximate the correlation between results from treatment periods, to allow inclusion of these studies in future reviews or meta-analyses.

Future studies might also address some issues in relation to the participants, which might also improve the certainty of findings. A greater number of participants are required in the studies. The level of ability/disability of the participants should be clearly defined using the most current classifications. For example, if it were used, the studies should focus on one level of ID or differentiate between the levels of ID when reporting the findings.

Future updates to this review should consider dividing it into more focused reviews, specifically for the four main interventions: comparing special manual toothbrushes, electric toothbrushes, training of carers of people with ID and training of people with ID. Additionally, future updates might consider excluding NRS from the reviews of these interventions; as it is now clear that RCTs exist and that the findings of the NRS tend to follow the same direction as the RCTs.

To more carefully assess any changes in the behaviour of participants that might occur within an intervention, the use of the COM-B framework, to identify

and reflect on the contexts and mechanisms of action at play in the intervention, or the consideration of other methods to measure outcomes of behavioural change, may be useful.

To allow valid comparisons between future studies, agreement in relation to the ideal index to use for the various clinical outcomes should be reached, such as a 'core outcome set', as recommended by the COMET initiative (Williamson *et al* 2017) and currently being developed by Lamont *et al* (2017) and Schwendicke *et al* (2015), in relation to periodontal disease and dental caries outcomes.

The clinical relevance of the findings from this review is unclear. There is much discussion regarding what reduction in gingival inflammation and plaque must be achieved to be clinically relevant, some levels of both conditions are likely to be present in even the healthiest of individuals. Plaque on less than 20% of the teeth is the target generally aspired to by oral care professionals, for their patients; a consistent plaque score of between 20% and 30% and a GI score of 2 or less has been shown to result in tooth survival in adults over a period of 26 years (93.8%) (Lang *et al* 2009). However, the baseline levels of gingival inflammation and plaque in the majority of the studies included in this review were much higher than those of data from surveys of general populations. Even the greatest reductions, reported in the interventions, did not reach either of the targets outlined above. Whether any reduction is better than no reduction is unclear. What is clear, is that the reductions must be sustained over many years for clinical benefits to be achieved (Lang *et al* 2009).

Future studies should consider including an assessment of dental caries, quality of life changes or long-term, patient-centered outcomes. The length of the follow-up period would thus need to be extended in many instances. Long-term follow-up for all of the outcomes, to show if any short- and medium-term benefits are sustained, is important. Finally, study registration and improved reporting would improve the ratings of risk of bias found in this review.

To conclude, the findings from this review contribute to the evidence available to healthcare professionals, when making decisions regarding the advice they

give to people with ID, and their carers, in relation to the type of toothbrushes they should use to improve the oral hygiene of people with ID and the training of value to them.

Nothing can be done to improve the quality of the studies included in this review. However, this review may contribute to the quality of future oral health studies for people with intellectual disabilities, by guiding those designing and reporting oral health interventions, in relation to the recommended duration of the intervention, the outcomes to assess and the indices used to assess them as well as the type of data to report.

Chapter 5 A Realist Review

What is it about carer-led oral hygiene interventions for people with intellectual disabilities that works and why?

5.1 Introduction

The oral health of people with more severe levels of ID is heterogeneous and often poorer than those with less severe levels of ID (Gabre *et al* 2001, Crowley *et al* 2005, Whelton *et al* 2009, Anders & Davis 2010, Petrovic *et al* 2016). People with moderate, severe or profound intellectual disabilities may require assistance and/or another person to carry out routine oral hygiene measures for them.

Oral hygiene interventions targeting the carers of people with more severe levels of ID are particularly complex, as the primary outcome of improved oral hygiene of the person with ID is dependent on the secondary outcome of engagement by the carer. This complexity makes Realist methodology the perfect choice to unravel some of these complexities and their impact on the outcomes of interventions, as it combines quantitative and qualitative evidence, resulting in a deeper synthesis and allows theories to be generated in relation to how and why an intervention does or does not work.

The focus of this Realist Review, as outlined in Chapter 2, was limited to carer-led oral hygiene interventions for people with ID, which were defined as: any carer-led/carers-implemented intervention that may impact on the duration, frequency and/or effectiveness of oral hygiene practices for people with intellectual disabilities or their carers. The decision regarding the focus of the Realist Review was made over a number of months, based on the early findings of the Cochrane Review, in relation to the extensive role played by carers in providing oral hygiene care, particularly in relation to people with severe or profound levels of ID. Of the 34 studies included in the Cochrane Review, 31 studies involved active input from the formal and/or informal carers in the intervention or they themselves were the focus of the intervention. The carer input included playing an active role in the toothbrushing routine of people with ID (assisting or fully undertaking the toothbrushing) or supporting, encouraging or monitoring the toothbrushing routine of people with ID. The

interventions focused on the carers themselves involved their education and training in oral hygiene theory and/or skills and/or behavioural change techniques. The impact of carers engagement with an intervention much be acknowledged.

The decision regarding the focus of the Realist Review was supplemented with methodological guidance from attendance at the CARES Realist training courses and workshops, in relation to the likely amount of relevant data and the time limitations for the review, as well as support from a local Realist Research Study Group (Trinity College and University College Dublin).

5.1.1. The aim of the Realist Review

The aim of the Realist Review was to identify the contexts and mechanisms that influence the outcomes, implementation and sustainability of carer-led oral hygiene interventions for people with intellectual disabilities.

The protocol for the Realist Review was submitted to Prospero (April 2018) and accepted and published in May 2018 (Registration Number CRD42018088163).

5.2 Methods

The steps of the Realist Review are broadly outlined in a process diagram which was developed as a process guide for the team, based on the RAMESES training manual (Figure 32) (Wong *et al* 2013). Each step will be explained in this chapter under the headings numbered 1 – 5 and their component parts.

A Realist Review is an iterative process; reflection on the process and findings from one step will influence the direction taken in the next, and so on. It was important while undertaking this Realist Review, to record the reflections and decisions made at the various stages so that a transparent record of the review process was created. This is to help the reader understand how the decisions were reached and to allow them replicate or amend the process if so desired.

The presentation of this chapter will, therefore, present the methods and findings of each step together, to provide the reader with an understanding of why a particular direction was taken and at what step in the process.

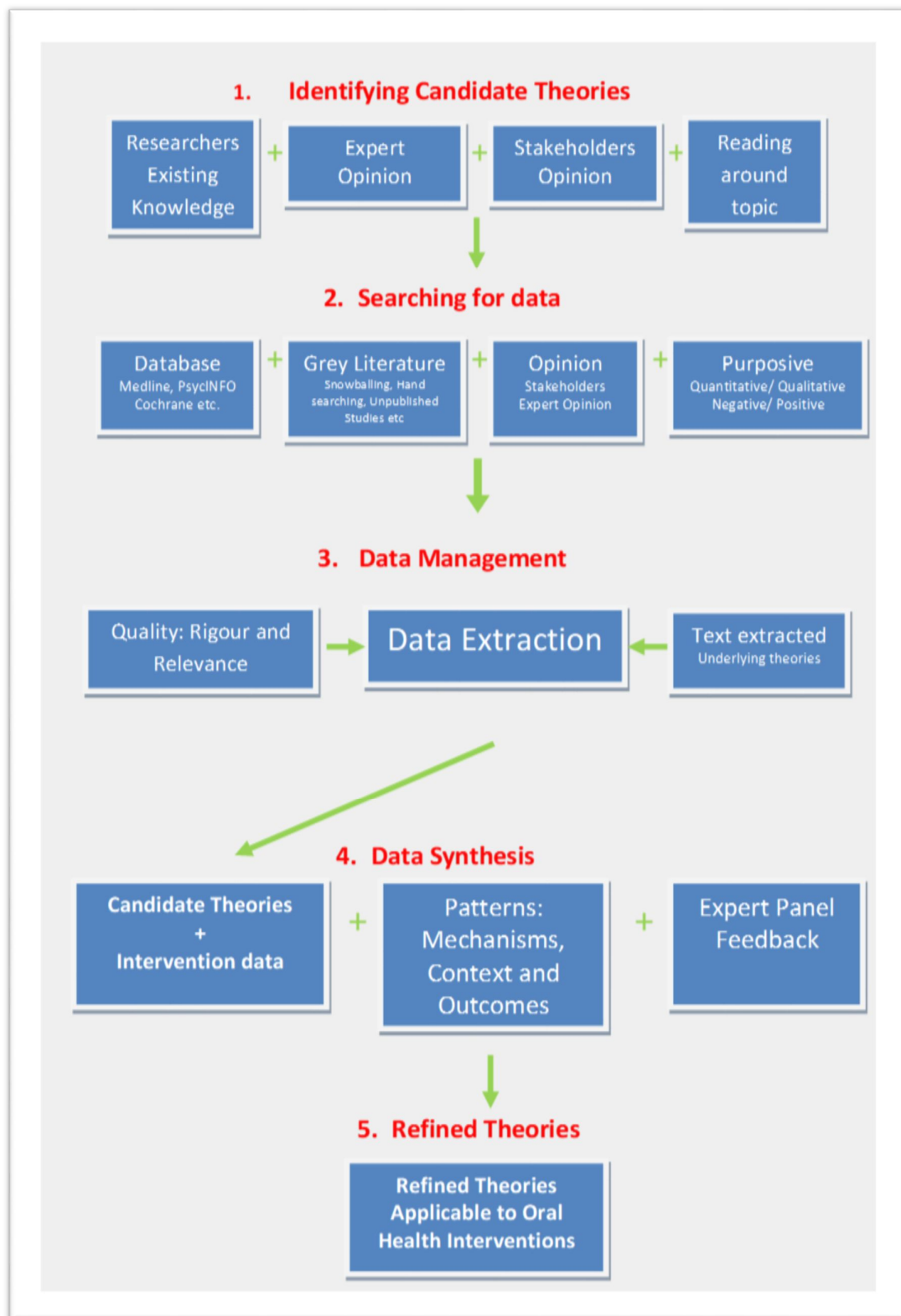


Figure 32: Realist Review steps

5.3 Step 1. Identifying Candidate Theories

Having determined the focus of the Realist Review, the first step was to develop the 'Candidate Theories' in relation to how carer-led oral hygiene interventions are supposed to work. Candidate theories' are the set of theories developed by Realist researchers at the outset of a Realist Review, based on their existing knowledge and a brief review of the literature. Throughout the Realist Review process, the Candidate Theories will be compared against the data identified from the search of relevant studies, to determine if the data confirm or refute the theories or if the theories require refinement.

Reading around the topic

Realist Research Standards for the full Realist search were followed and go beyond what has already been completed for the earlier reviews in this thesis. The search will be detailed later in this chapter. For this preliminary stage of the process, the search for relevant information to develop the Candidate Theories began with reading, with a Realist lens, some relevant studies, which had been identified from the Scoping Review reported earlier (Waldron *et al* 2016). A broad search was then undertaken to find some specific carer-led oral hygiene interventions. These interventions were identified from a PubMed search using the terms listed below and some citation and forward and backward searches on the findings:

TITLE-ABS-KEY ("carer-led" OR "caregiver-led" OR "carer-assisted" OR "caregiver-assisted" OR "carer-directed" OR "caregiver-directed" OR "parent-led" OR "parent-assisted" OR "parent-directed" OR "spouse-led" OR "spouse-assisted" OR "spouse-directed") AND oral hygiene AND ("intervention" OR "program" OR "review").

This search also identified some relevant carer led, non-oral health, chronic health interventions, which it was felt might produce some relevant theories. The purpose at this stage was to identify what were felt to be the facilitators and barriers that may influence the outcomes.

In the process of reading the reports of the interventions, familiarisation with the concepts of mechanisms (how people interpret or act upon an intervention)

and contexts (the elements that may affect the activation of the mechanism) as defined by Pawson & Tilley (2004), was undertaken by the review team. Following the reading of a randomly selected number of the reports, a brainstorming session was held by the review team to discuss the details with a view to developing the candidate theories. This process involved the team members independently identifying what they felt were relevant contexts and mechanism in the studies and recording how they might impact on the outcomes. This was followed by comparison of the context and mechanisms identified, with discussion regarding their relevance to the research questions.

The review of the literature on the formal theories of behavioural change used in oral health interventions, which was undertaken at the outset of the Scoping Review, and presented in Chapter 1, Section 1.7 (Pages 17 – 26) of this thesis, was also helpful in the process of developing the candidate theories.

Researchers existing knowledge

It is recommended that the review team have an expertise in the topic being reviewed (Wong *et al* 2013). This ensures that the 'Candidate Theories' developed at the beginning of the process are relevant to the topic being reviewed. My background is in oral health promotion, communication and motivation in the oral healthcare setting. Having some experience in both general and oral health promotion of the general population, I come to this review with some preconceived ideas and knowledge of the formal theories in relation to how health interventions for the general population are likely to work. Prof. June Nunn and Dr. Caoimhin Mac Giolla Phadraig (the other Realist Review team members), are specialists in Special Care Dentistry, with vast experience in providing oral health care for people for ID and in advising their carers.

Stakeholder opinions

Development of the candidate theories is an important phase of the Realist Review: it may dictate the direction taken in the review. As I have no direct experience with implementing interventions focused on people with an ID, I felt it was important to get a better understanding of the issues likely to be encountered. The input of others, who have actually implemented or been part

of a relevant intervention might provide valuable insight to inform the development of the candidate theories. For this reason, I undertook some interviews with dental professionals who have experience in this field, whom I termed "Local Experts". The interviewees included special care dentists, general dentists with an interest in Special Care Dentistry, an oral health promoter and a dental nurse. The interviewees worked in both the private and public oral health sectors in Ireland.

The interviews were prefaced with an introductory email (Appendix 8) detailing the project and a request for their input either by completion of a prepared form (Appendix 9) or by interview. The email was sent to twelve people, two were undelivered, and responses were received from nine. Six of these were interviewed by phone and one face to face, two of these also completed the written form. The remaining two were not interviewed; after completing seven interviews I felt I had sufficient data to help me develop candidate theories. As further feedback would be required in the future, I felt it would be sensible to keep these experts for these later stages.

The interviews took place over a period of two weeks in February 2017. All the telephone interviews lasted approximately 30 minutes and the face to face interview lasted almost an hour. The interviews were broadly structured using the prepared form (Appendix 9), which looked for details of the interventions that the interviewee had experiences of, the facilitators and barriers they felt impacted the outcome and finally, any general thoughts on the topic.

The interviews were recorded, with the permission of the interviewees, to ensure accurate records and retention of the verbatim phrases used by the interviewees when describing their experiences.

The Local Experts appeared to easily understand the purpose of the interview and were able to provide examples for all elements outlined on the prepared form. Many were able to give details of specific oral health interventions with which they had been involved, and all were able to give details of one-to-one interactions with people with ID and their carers. This direct contact with local experts was particularly useful in identifying some of the mechanisms that may

impact on the outcomes of interventions, which are not often specifically reported or referred to in published studies.

The review of the literature from the Scoping Review and the information received from the Local Expert's interviews provided a picture of what carer-led oral hygiene interventions for people with intellectual disabilities generally look like. Interventions most commonly focus on education, skills training and encouragement of carers to undertake regular oral hygiene care routines. The formal theories underlying the interventions are often vaguely referred to or missing altogether. Qualitative data, such as how and why the interventions worked, or not, are often only referred to in the discussion section of the published studies.

Themes that emerged in relation to explaining poor outcomes refer to the difficulty of timing the training to suit the carers, the specificity of the training, the issues of rosters and movement of carers, the lack of support or clear policy from management and other staff, and the lack of engagement of carers with oral health issues in general.

Expert Knowledge

My involvement in the Realist Research Study Group was helpful in relation to the construction of the candidate theories based on the information gathered. Each group member took turns to present their candidate theories to the group, it was interesting to observe that individual group members used slightly different methods to identify the contexts, mechanisms and outcomes. However, for all of us, it involved steeping ourselves in the literature; reading and re-reading the studies; periods of reflection; and refinement of the terms being used. Some group members were more visual and needed to draw diagrams and flow charts, others, like me, were more structured using tables with the key elements identified.

The terms used to describe mechanisms vary from one researcher to another. Pawson suggested, at a Realist Expert workshop and conference I attended to help in the development of the candidate theories (2016), that "Mechanism Libraries" should be developed so that mechanisms can be grouped, in order

that generalised theories can be developed. For example; “This class of mechanism will tend to act on this class of subject in this class of circumstances”. We are some way away from this yet, however, Pawson’s other suggestion, that Michie’s library of behaviour change systems was a good place to start, proved useful to me (Michie *et al* 2013, Pawson 2016). Importantly, I found I needed to clearly define the terms I was using to verbalise my possible mechanisms, at the time that I was categorising them, to avoid confusion at later iterations.

Drafts of Candidate Theories

Alongside the interviews and study group sessions, I also used QSR International's NVivo 11 software to identify the contexts, mechanisms and outcomes (CMOs) in the text of the studies identified in the initial search, as well as from my summary notes of the recordings from the local experts’ interviews (NVivo 11 2012). This software is particularly helpful in managing the CMO’s from all the reviewed studies, allowing patterns to emerge from across the studies, while still maintaining the exact sources of the CMO’s.

The first, embryonic, draft of the candidate theories was based on full reading of all the selected studies, following the pilot of the process and brainstorming with the Realist Review team (Table 59). Subsequently, a form for extracting theories from the studies, and a new format for outlining the theories were developed, based on knowledge gained from discussions with the Realist Experts, the Realist Review team and the Realist Research Study Group and deepening familiarization with earlier realist research carried out by respected realist researchers such as McMahon & Ward (2012) and Pearson *et al* (2012, 2015). These researchers provide detailed documentation of the processes followed in their reviews which is broadly outlined in the diagram by McMahon & Ward (Figure 33).

Table 59: First Draft of Candidate Theories

(based on a small selection of relevant studies using NVivo with CMOs identified as **Context** (Bold) Mechanism (underlined) *Outcome* (italics)

a)	Carers engagement is essential for <i>oral hygiene practices for people with ID to be maintained</i>
b)	Tailored educational material to suit the educational level of carers will improve <u>engagement, knowledge and attitude</u> of carers
c)	Tailored skills training relative to the level of ID and/or cooperation of the client, will improve <u>self-efficacy</u> of the carer, thereby <i>increasing the level of oral hygiene practices</i> undertaken by the carer
d)	<u>Support</u> from other professionals such as dental and general health care professionals and teachers will assist long term <u>engagement</u> by the carers, in <i>oral hygiene practices</i>
e)	<u>Flexibility</u> in relation to access to training for carers will <i>increase participation in oral hygiene practices</i>
f)	Repeat training will <i>increase skills level and confidence</i> of carers to undertake oral hygiene practices
g)	Incorporation of oral hygiene practices into the daily schedules of clients will improve levels of formal carer <u>engagement</u> thereby <i>improving levels of oral hygiene practice</i>
h)	Support from management will improve levels of <u>engagement</u> of formal carers in <i>regular oral hygiene practices for the people they care for</i>

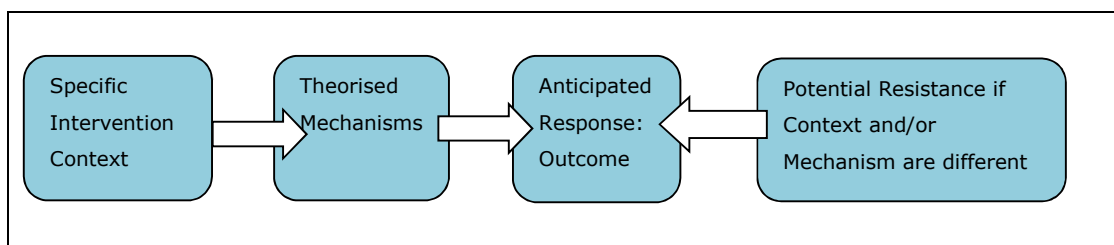


Figure 33: Intervention Change Chain (McMahon & Ward 2012)

Significant redrafting, development and iteration of the theories took place during this process, with input from the Realist Review team, the Realist Experts and one of the Local Experts, not used in the earlier interviews, who read and gave feedback on the theories in relation to the clarity, relevance and

ability to implement them in practice. The final drafts of the ten candidate theories are outlined in Table 60 (Pages 211 – 221).

Table 60: Revised Draft of Candidate Theories

Theory title	Context	Resource Mechanism	Mechanism Response	Outcome	Theory – context, mechanism, outcome	If... then statement	Barriers
Theory 1: Engagement	Lack or low level of knowledge and understanding of the consequence of poor oral health for people with intellectual disabilities (ID), resulting in lack or low level of interest in the oral health of people with ID	A training opportunity that attracts carers to get involved in oral hygiene for people with ID	Enticement, Engagement, Motivation, Initiation Curiosity	Attractive training opportunities will ensure engagement and motivation of the carers to attend training, initiate and maintain salutogenic behaviours in oral hygiene for people with ID	Carers limited knowledge and understanding of the consequences of poor oral hygiene results in their poor engagement in oral hygiene practices for people with ID. <u>Carers must be provided with attractive training opportunities that result in positive engagement with the training and increased motivation to initiate and sustain delivery of oral hygiene practices for people with ID.</u>	If Carers are provided with attractive training opportunities, then there will be increased attendance and engagement in training resulting in increased knowledge and motivation to initiate and sustain the delivery of oral hygiene practices for the people with ID for whom them care.	Lack of engagement by the carer will result in poor levels of attendance at training and poor levels of initiation of OH care practices for the people with ID for whom they care.

Theory title	Context	Resource Mechanism	Mechanism Response	Outcome	Theory – context, mechanism, outcome	If... then statement	Barriers
Theory 2: Systems Level Approach	Current government policy, health service policy, Quality control of oral health services, local settings policy, level of support from management and work colleagues, training of health care workers in relation to oral health	Oral hygiene interventions planned at the systems level	Embed values, Establish norms, Accountability, Motivation, Self-confidence, Incentivisation	Establish values and norms, and understanding in relation to the OH care of people with ID among all stakeholders in the system	Oral hygiene interventions should be planned at the systems level <u>in order to embed values into management practices, thus ensuring oral hygiene practices for people with ID become established as norms throughout the organisation, for which people are accountable, at all levels.</u>	If interventions to establish daily oral hygiene practices for people with ID are planned at policy and management level and embedded into management practices, then these practices will be established as norms throughout organisations and people will be accountable for these practices at all levels.	Lack of support and understanding from other stakeholders will result in poor sustainability of the intervention by Carers.

Theory title	Context	Resource Mechanism	Mechanism Response	Outcome	Theory – context, mechanism, outcome	If... then statement	Barriers
Theory 3: Local Policy	Local policies and guidelines - particularly in relation to consent /assent and autonomy	Awareness of local policies and guidelines in relation to interactions with people with ID when designing and implementing OH interventions	Clarity, Assurance, Self-confidence, Engagement Duty	Awareness of local policies and guidelines in relation to interactions with people with ID when designing and implementing OH interventions will give clarity, assurance, a sense of duty and self-confidence to the carer and increase engagement in relation to their OH role.	<u>Awareness of local policies and guidelines when designing and implementing interventions will provide the opportunity to avoid conflict or confusion and increase the engagement and self-confidence of the carer in providing oral hygiene care for the people with ID for whom they care within their setting.</u>	If intervention planners take into consideration the local policies and guidelines that are already in place for carers e.g. in relation to consent or autonomy, then this will avoid conflict or confusion in relation to how the intervention will work in their setting, providing clarity, assurance and self-confidence to the carers and increasing their engagement with the planned intervention.	Carers will lack confidence, direction; become confused and disengage in the intervention if local policies and guidelines have not been considered when designing and implementing the intervention.

Theory title	Context	Resource Mechanism	Mechanism Response	Outcome	Theory – context, mechanism, outcome	If... then statement	Barriers
Theory 4: Tailoring to the person with ID	Each person with ID has different needs in relation to assisting them or undertaking their daily oral hygiene. The resources available for each person with ID and the daily routines of the person with ID and the carer are different.	Skills Training for Carers that includes tailoring for the environmental and personal needs of the person with ID for whom they care	Awareness Self-confidence, Self-efficacy, empathy, compassion, adaptability	Increased ability of the carer to adapt to the needs of the person with ID will result in a more tailored approach to their OH care, benefiting both the person with ID and the carer physically and psychologically.	<u>Training for carers that includes tailoring to the environmental and personal needs of the person ID for whom they care will increase the carers potential to respond to individual needs and thus their ability to adapt to the context of the person with ID; improve the carers self-efficacy and self-confidence,</u> thereby <i>increasing the likelihood of successfully providing sustained oral hygiene care for the person with ID.</i>	If carers are trained to tailor the oral hygiene care they provide to the personal and environmental needs of the individual person with ID for whom they care, then their levels of self-efficacy, and self confidence in adapting and providing this sustained care will increase.	Failure to take into consideration the environmental and personal needs of the person with ID when undertaking OH practices for them, may result in physical and psychological stress of both the carer and the person with ID leading to poor sustainability of the practice.

Theory title	Context	Resource Mechanism	Mechanism Response	Outcome	Theory – context, mechanism, outcome	If... then statement	Barriers
Theory 5: Tailoring to the Carer of people with ID	The routines and other commitments of the carers. The attitude of the carer to oral health. The understanding of the impact of daily oral hygiene on the health of people with ID. The feelings of fear or disgust when undertaking oral hygiene for people with ID. Access to training. Support of other (oral) health professionals.	Training for Carers that considers their environmental and personal contexts	Flexibility, Understanding, Normalisation, Engagement, Self-confidence, Self-efficacy, Motivation	Provide accessible and tailored training to Carers to meet their needs, allowing greater understanding and normalisation of the OH care needs of people with ID, increasing the likelihood of sustained engagement in providing OH practices for people with ID.	<u>Training for carers that includes tailoring to their environmental and personal contexts, will provide opportunities for flexibility, increased understanding, normalisation, engagement, self-confidence, self-efficacy and motivation</u> , which will <i>increase the likelihood of successful training and undertaking of sustained oral hygiene practices for the people with ID for whom they care.</i>	If the training provided to carers is tailored to their personal and environmental contexts and needs, then their engagement with this training will increase, resulting in an increased understanding and normalisation of the elements that can cause fear or disgust, and increased knowledge, self-confidence, self-efficacy and motivation to undertake sustained oral hygiene practices for the people with ID for whom they care.	Assuming all carers are equal, lack of appreciation of individual constraints and capacity

Theory title	Context	Resource Mechanism	Mechanism Response	Outcome	Theory – context, mechanism, outcome	If... then statement	Barriers
Theory 6: Reinforcement	Level of support from management and other staff, Level of support to non-formal carers from health services. Feedback from efforts made to improve oral hygiene of the people with ID for whom they care. Level of inclusion of oral hygiene/ oral health in general health assessments of the people with ID.	Support from other staff, outcome reviews and positive feedback regarding OH care of people with ID undertaken by carers provides reinforcement to continue their efforts.	Support, Encouragement, Motivation	Positive feedback will provide encouragement and motivation to the carers to continue providing OH care for the people with ID for whom they care.	<u>Reinforcement, from outcome reviews, support from other staff and positive feedback, of the carer-delivered OH practices for the people with ID will provide encouragement and motivation to the carers to continue delivering the daily oral hygiene practices for the people with ID for whom they care.</u>	If support and positive feedback is provided to carers in relation to the oral hygiene care they provide to the people with an intellectual disability for whom they care, then they will feel encouraged and motivated to continue providing this care.	Carers will lose the motivation to continue their efforts, if they are not aware of the positive effects they are having on the health of the people with ID for whom they care.

Theory title	Context	Resource Mechanism	Mechanism Response	Outcome	Theory – context, mechanism, outcome	If... then statement	Barriers
Theory 7: Goals	Level of monitoring of oral hygiene routines for PID. Lack of clear steps in the oral hygiene routine. Lack of Incentivisation or repercussions if the oral hygiene routine is not completed.	Agreed goal setting, in relation to OH practices for PID, with rewards or incentivisation, by Carers and other stakeholders (i.e. PID, trainers, care staff, families, management).	Intention/ Goal, Incentivisation, Motivation, Encouragement	By setting agreed goals, with other stakeholders, the Carers will be incentivised to undertake and continue providing oral hygiene care to the people with ID in their care.	<u>Agreed Goal setting with achievable steps, will provide the carer with a reminder of their intention and an incentive to undertake daily oral hygiene care for the people with ID.</u>	If goals, rewards or incentivisation and achievable steps are agreed with carers in relation to providing daily oral hygiene care for the people with ID for whom they care, then they will have an incentive and a reminder of their intention to provide this care.	A lack of clear goals for the Carer may result in a loss of direction and motivation to achieve the end objective.

Theory title	Context	Resource Mechanism	Mechanism Response	Outcome	Theory – context, mechanism, outcome	If... then statement	Barriers
Theory 8. Professional Support	New clients/PID, new issues, Carers lack of knowledge in relation to dealing with issues such as resistance to care, access to areas of the mouth, positioning of the carer and PID when carrying out OH, oral health conditions	Professional advice, resources, aids, encouragement, skills training, solutions	Support, Encouragement Self-efficacy, Self-confidence and engagement of carers	By providing professional support when needed, carers will be able to find solutions to deal with any barriers to providing the OH care, resulting in sustained oral hygiene care for the people with ID.	<u>Support from professionals will provide opportunities to discuss the barriers to providing oral hygiene care and identify solutions, which will increase self-efficacy and self-confidence and assist long term engagement by the carers, in sustained oral hygiene practices</u>	If carers can access professional support for problems or barriers that arise when providing oral hygiene care to PID, then they can find solutions to these barriers, increasing self-efficacy and self-confidence and resulting in continued engagement in providing sustained oral hygiene practices for the people with ID for whom they care.	Absence of support and guidance will cause programmes to falter/fail

Theory title	Context	Resource Mechanism	Mechanism Response	Outcome	Theory – context, mechanism, outcome	If... then statement	Barriers
Theory 9. Regular Knowledge and Skills Training	Fading of knowledge and skills, staff movement - leave, rotations etc., new staff, staff turnover	New, updated or revised knowledge and skill training	Increased knowledge, Self-efficacy, Self-Confidence, Motivation, Engagement	Clinical signs of gingival disease (MGI/ GI) or dental caries (DMFT), Plaque levels, adherence to oral hygiene routines, oral hygiene practice measures, oral assessment, carers self-efficacy scales, changes in attitude and knowledge of carers.	Staff turnover and/or rotation and fading of knowledge means that <u>knowledge and skills training should be repeated on a regular basis, this regular training will result in increased levels of engagement, motivation, self-efficacy and self-confidence in carers providing oral hygiene care practices for the people with ID for whom they care, impacting positively on their oral health.</u>	If regular knowledge and skill training is provided to carers then issues such as staff turnover and knowledge fading will not impact on the levels of engagement, motivation, self-efficacy and self-confidence of the carers, and they will continue to provide sustained oral hygiene care practices for the people with ID for whom they care.	Lack of prioritization for appropriate resourcing for regular training

Theory title	Context	Resource Mechanism	Mechanism Response	Outcome	Theory – context, mechanism, outcome	If... then statement	Barriers
Theory 10 Personnel involved	Level of stakeholder's involvement, oral health care professional, medical professional, psychologist, teachers, families, people with ID. Different emphasis or focus by different stakeholders. Multi-stakeholder team.	Interventions designed by multi-stakeholder teams will provide a more rounded intervention Vision in place	Supported, involved, expertise recognised, appreciated, rounded approach, organised, complete	Increase the sustainability of the intervention	<u>A multi-stakeholder team who have</u> designed, implemented and evaluated an intervention <u>will feel supported and appreciated, have considered all of the complexities and be organised and prepared to deal with any issues,</u> which will <i>increase the sustainability of the intervention.</i>	If the intervention is designed, implemented and evaluated by a multi-disciplinary team, then any issues arising will be dealt with without recrimination, in a supportive way, thereby increasing the sustainability of the intervention	Fragmented approach may lead to disillusionment and lack of adherence to intervention

Expert Panel review of the Candidate Theories

Once the candidate theories had been finalised, they were sent to a panel of international experts in special care dentistry, all of whom have researched and published in this area, to review and provide feedback, based on their own knowledge and personal experience in the field: this group was termed the “Expert Panel” (Table 61). The panel members were recommended for inclusion on the panel by two of the Realist Review team members who are special care dentists and are involved professionally at an international level. The theories were sent to the Expert Panel in Survey Monkey format along with a background information sheet to explain the Realist method and the purpose of the exercise (Appendix 10). The panellists were given the opportunity to rate each theory in relation to clarity, relevance and ability to implement it in practice as well as a request to provide general comments.

Table 61: Expert Panel members

Expert Panel Member	Work /Expertise	City, Country
Archana Pradhan	Senior Lecturer in Dentistry, University of Queensland. Fellow of Special Care Dentistry. Lead for People with Disabilities, Australia’s National Oral health Plan (2014 – 2023)	Adelaide, Australia
Johanna Norderyd	Specialist in Paediatric Dentistry at the National Oral Disability Centre for Rare Disorders. She has a PhD in Disability Research and is a member of the Executive Board of the International Association for Disability and Oral Health.	Jonkoping, Sweden
Lorna McPherson	Professor of Dental Public Health and Co-Director and Evaluation Lead of the national Childsmile oral health improvement programme UK. A member of the Scottish Oral Health Research Collaboration and a major player in the development of research related to improving the oral health of older people.	Glasgow, Scotland
Catherine Binkley	Associate Professor, Hospital Dentistry, University of Louisville. Research focusing on interventions with vulnerable and other special care patients.	Kentucky, USA

Imke Kaschke	An independent researcher/dentist focusing on athletes in the Special Olympics. Formerly Head of the Specialized Dental Clinic for Disabled Persons, Medical School - Charité - University Medicine Berlin and Lead, Master programme Public Health, Freie Universität Berlin.	Berlin, Germany
Dominique Declerck	Chair of Population Studies in Oral Health and head of Department of Oral Sciences, Faculty of Medicine University of Leuven. Research focus on dental public health, dental epidemiology and statistics.	Leuven, Belgium
Conac Bradley	General Dentist, senior dental officer in Health Service - Special Care Dentistry. Past President, Irish Society of Disability and Oral Health.	Wicklow, Ireland
Shelagh Thompson	Professor, School of Dentistry University of Liverpool. UK Honorary Consultant in Special Care Dentistry, Royal Liverpool and Broadgreen University Hospitals NHS Trust. Past Editor, Journal of Disability and Oral Health	Liverpool, UK

The final versions of the candidate theories were shortened to improve their comprehension for people unfamiliar with Realist methodology and terminology, following discussion with the team and a pilot with one of the local experts, and in no particular order, were sent to the expert panel as follows:

Theory 1: Oral hygiene interventions planned at the systems level, embedding values into management practices, will ensure that oral hygiene practices for people with intellectual disability become established as norms throughout the organisation, encouraging people at all levels to be accountable.

Theory 2: When designing and implementing interventions, a familiarity with local policies and guidelines will avoid conflict or confusion and increase the engagement and confidence of carers when delivering oral hygiene practices for people with intellectual disability.

Theory 3: Carers must be provided with training that is meaningful and attractive to them resulting in their positive engagement and increased

motivation to initiate and sustain delivery of oral hygiene practices for people with intellectual disability.

Theory 4: Training for carers that takes into consideration the environmental and personal needs of people with intellectual disability will increase the carers' potential to adapt principles to individual needs, thus increasing the likelihood of them initiating and sustaining delivery of oral hygiene practices for people with intellectual disability.

Theory 5: The personal needs and environment of carers must be considered when planning their training, providing opportunities for flexibility and increasing the likelihood of successful training and of them initiating and sustaining delivery of oral hygiene practices for people with intellectual disability.

Theory 6: Positive feedback from outcome reviews and support from other staff will provide encouragement to and aid motivation in the carers to continue delivering oral hygiene practices for people with intellectual disability.

Theory 7: Agreed goal setting with achievable steps, will provide carers with a reminder of their intention and an incentive to undertake sustained delivery of oral hygiene practices for people with intellectual disability.

Theory 8: Support from oral care professionals will provide opportunities to discuss the barriers and identify solutions to providing oral hygiene care for people with intellectual disability, increasing self-efficacy, self-confidence and long-term engagement by the carers in sustaining delivery of oral hygiene practices for people with intellectual disability.

Theory 9: Staff turnover and/or rotation and fading of knowledge means that knowledge and skills training should be repeated on a regular basis, maintaining levels of engagement, motivation, self-efficacy and self-confidence of carers to sustain delivery of oral hygiene practices for people with intellectual disability.

Theory 10: A multi-stakeholder team who have designed, implemented and evaluated an intervention will feel supported and appreciated, will have considered all of the complexities and be organised and prepared to deal with any issues, which will increase the sustainability of the intervention.

5.4 Step 2: Searching for data

Once the feedback had been received from the Expert Panel, a more specific search of the literature was undertaken, in order to find evidence to refine the theories and explain the patterns, as outlined in the process diagram from the RAMESES training manual (Figure 34), (Wong *et al* 2013).

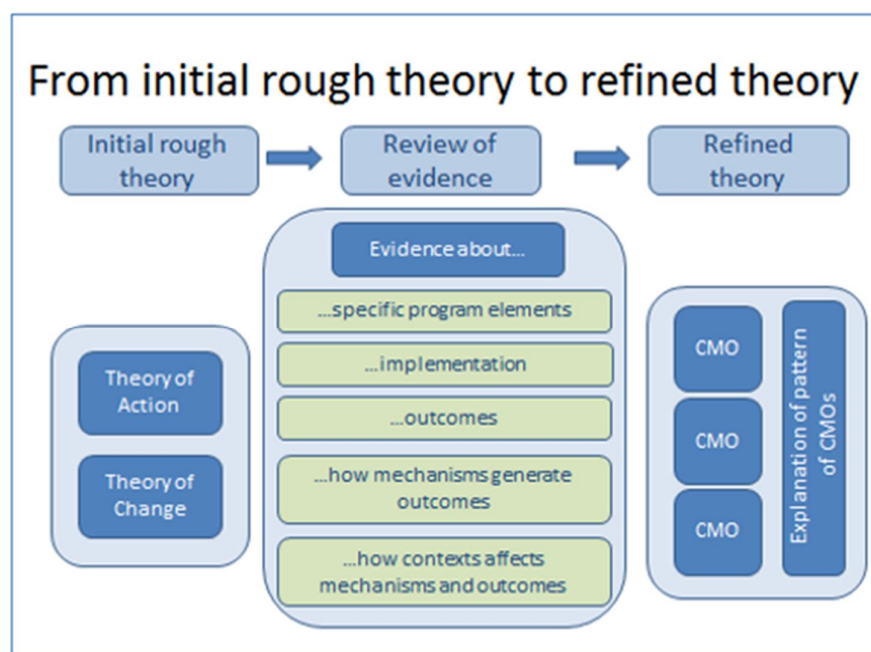


Figure 34: Realist theory refining process diagram (Wong *et al* 2013)

Search strategy for the Realist Review

The search strategy for a Realist Review needs to be more creative, intuitive and iterative, in order to uncover what is happening as a result of the intervention (Booth *et al* 2013). Often the detail of the conceptual theory of an intervention is not reported or reported separately to the details of the intervention itself; these details may not be uncovered in a conventional search. This iterative searching can result in a lack of reproducibility, thus detailed records and transparent reporting of the method used is essential. The search strategy should be as wide ranging as time and resources allow.

Search of the Databases

The search findings of the Cochrane Review of oral hygiene interventions for people with intellectual disabilities was used as a starting point. Both randomised controlled trials (RCT) and non-randomised studies (NRS) were included in the Cochrane search. This dual approach, of completing a systematic review alongside a Realist Review and using the search findings of the systematic review to inform the Realist Review, was novel when first outlined to the Realist Experts at a workshop in 2015, however, it has since been used in a small number of published Realist Reviews and considered a particularly good approach for reviews of complex interventions (Brown *et al* 2016).

The Cochrane Review, using the search strategy outlined in Chapter 4, Section 4.4.2 and in Appendix 1, searched the following electronic databases and trials registries: Cochrane Oral Health's Trials Register; Oral Cochrane Central Register of Controlled Trials (CENTRAL) in the Cochrane Library; MEDLINE Ovid (from 1946 onwards); Embase Ovid (previous six months to date); PsycINFO Ovid (from 1806 onwards), US National Institutes of Health Ongoing Trials Register ClinicalTrials.gov (ClinicalTrials.gov/); World Health Organization International Clinical Trials Registry Platform (apps.who.int/trialsearch). Hand searching of the specialist conference abstracts from the International Association of Disability and Oral Health (2006 to 2016) was also completed.

Purposive Searching

The Cochrane Review search resulted, after arbitration, in 178 relevant articles, at the title and abstract stage. For the Realist Review, 41 of these were excluded as they were in languages other than English. the remaining 137 articles were further filtered for terms under three broad headings of Context, Mechanism and Outcome, in order to identify any reference to carers and/or any formal or informal behavioural theories used, resulting in 39 potentially relevant articles (Table 62).

Table 62: The search terms used to filter the Cochrane Review findings

CMO's	Search terms
Context	Carer, caregiver, parent, theor* (theoretical), setting, management, policy, guideline, stakeholder, multi, context
Mechanism	Support, Training, motivation, knowledge, monitor, (self) confidence, (self) efficacy, goal(s), incentive*, feedback, encouragement
Outcome	Continue, attitude, increase, adapt, sustain

Following the RAMESES quality standards for the search strategy, a broad range of sources that may go beyond the area of oral health or intellectual disability where the role of the carer might be considered, was also searched. We searched particularly for qualitative reports, interviews or surveys of carers. We also actively searched for reports of negative findings. These searches were limited to literature in the English language (Table 63) (Wong *et al* 2013).

Table 63: Realist Review search terms

Realist Review Search Terms
("Caregivers"[Mesh]) AND "Staff Development"[Mesh]) OR "Health Policy"[Mesh]) OR "Attitude of Health Personnel"[Mesh]) OR "Work Engagement"[Mesh]) OR "Motivation"[Mesh]) OR "Personnel Turnover"[Mesh]
"Carer-led" AND (theor* OR model* OR concept* OR framework*)
"Carer Engagement" AND (theor* OR model* OR concept* OR framework*)
"carer training" AND (barriers OR facilitator) AND (theor* OR model* OR concept* OR framework*)
"carer training" AND (barriers OR facilitator) AND "oral health" AND (theor* OR model* OR concept* OR framework*)

Grey Literature searching

Snowballing was used: this included searching the list of references in the included articles and using the "similar articles" and "forward citation searching" features on the databases, if available. The Web of Science list of articles citing Simons 2000 "An evaluation of an oral health training

programme for carers of the elderly in residential homes”, was a particularly fruitful search adding 44 additional articles for full text reading. However, only a small number of relevant grey literature sourced articles, not already identified from the early searches, were ultimately included in the review, suggesting that the search terms used for both the Cochrane and Realist searches were effective.

The Prisma flow chart outlines the results of the searches (Figure 35).

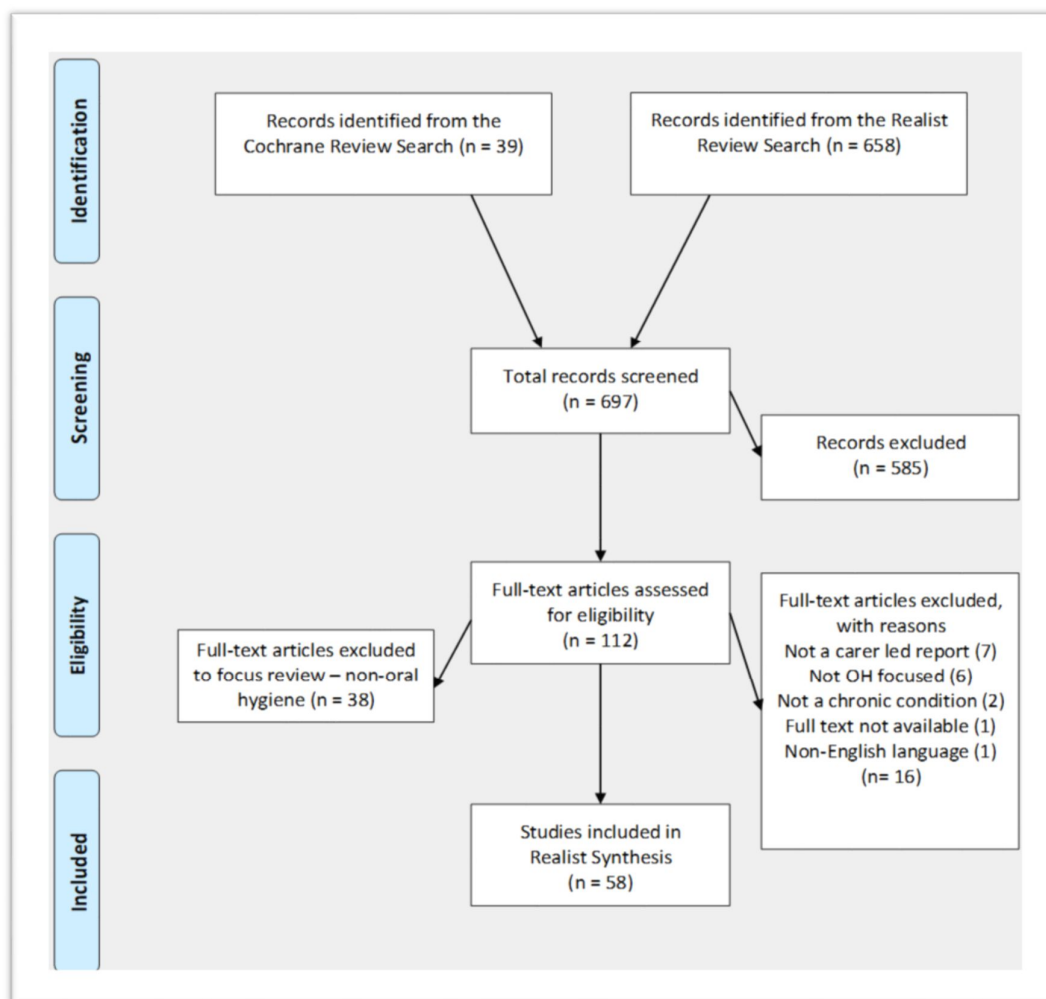


Figure 35: Prisma Flow Chart for the Realist Review

5.5 Step 3: Data Management

Title and Abstract Screening Criteria for Realist Review

Following the filtering process of the Cochrane articles, thirty-nine articles were identified as potentially having information relevant to the Realist Review. A

total of 658 articles were identified using the realist review terms. A total of 697 articles were screened by title and abstract, using the inclusion and exclusion criteria outlined below:

Inclusion criteria:

- Population - Formal or non-formal carers providing care on a continual basis for people with any chronic /long-term health condition.
- Type of Report - All report types are eligible including, for example, policy documents and opinion pieces, the report does not need to be an intervention.
- Relevance – Any report that suggests that there might be information of value to the review question/ our candidate theories – in particular, anything that might add information in relation to contexts or mechanisms which might influence carers behaviour.
- Rigour - the sources of the relevant information are credible and trustworthy.

Exclusion criteria:

- Not involving carer led care provided on a continual basis (i.e. not on a daily basis) or provided short term (i.e. caring for a person after a hospital visit) or terminal illness (i.e. cancer).
- Any report relating to carers levels of stress, mental health or burden of care, if not also reporting the impact on their provision of care.

Rigour and Relevance

A pilot screening of 10% of the articles was undertaken by the three review members to ensure the criteria were clear, being followed and were resulting in articles relevant to the review. Disagreements were explored and the criteria were clarified. The remaining articles were screened by me alone. From the 697 articles screened, 585 were excluded, leaving 112 articles for full text screening. Most of the full text of the articles were sourced from the DDUH and TCD libraries; eleven were sourced via inter-library loans.

As a means to assess the quality of the studies, rigour was also assessed throughout this full text reading process. Suggested methods to assess rigour

in Realist research include assessing issues such as: who is saying it; are they respected in their field, have they relevant experience, does it match what is being said by others who have confirmed rigour (from research) and does it make sense (Wong 2018).

Data Extraction

Data extraction was undertaken between October and November 2018. The 112 articles were uploaded to the NVivo 11 software package, becoming the "Sources" of data. Then folders were set up on NVivo to organise any extracted data, referred to as "Nodes", relating to the ten candidate theories. Reading of the full text and extraction of the data, into/linking to the relevant nodes, were carried out concurrently.

As a trial of the extraction process, the first ten articles in alphabetical order were read and sections of text were highlighted and linked to the ten candidate theory nodes, if they were felt to contain data relevant to the context, mechanisms or outcomes of the theories. Four additional nodes were set up during this process to capture; 1. references to other studies that might be relevant, 2. theories not considered as a "catch all" node for anything that came up that did not seem to fit into any of the existing ten nodes, 3. any formal theories of behaviour change identified in the studies and finally, 4. to summarise the outcomes of the studies. Reassuringly, it was noticed during this trial process, that many of the references to other, possibly relevant, studies within the articles were from studies already included in this review, suggesting that the search methods used were appropriate.

After reading and extracting data from the ten articles, while regularly re-reading the protocol and theories in order to keep my focus on the research questions, I reviewed the information extracted to see if it was relevant or could be recorded in a more efficient way. This was a useful process as I could see that I needed to capture the context around the data I was extracting, or it would not make sense to me when I reviewed it later. I was also tending to extract some data that were not relevant to the theories. The data that were relevant needed to be grouped; new sub nodes were set up within the main nodes, to capture separately the facilitators, barriers, mechanisms and

contexts. This helped to ensure the data extracted were relevant to the research questions and helped me to stay focused.

The need to focus the breadth of the review, after extracting data from ten articles, became obvious when taking into consideration the time it took and the time available to complete it, as well as the richness of the data being extracted. Options considered for focusing the review at this point included: limiting it to oral hygiene interventions only (this would include other populations but keep the outcomes similar), limiting it to those interventions that considered the oral health of the person with the chronic illness (exclude those interventions that are assessing the carers knowledge or attitude only) or limiting the articles based on their age, including the more recent articles only. After discussion with the review team, a decision was taken to focus on oral hygiene interventions only, whilst maintaining the range of populations with chronic conditions (People with ID, Elderly, Stoke patients, Patients with dementia, children and adults with non-specific disabilities).

Full text reading of the remaining articles was then completed. Articles were excluded if it became apparent that the focus was on supporting carer welfare, the medical condition was one that could not be compared to providing oral hygiene care on a regular basis or was one where the motivation of the carer was influenced by the medical condition, i.e. life limiting. Other reasons for excluding articles were that the focus was on dental treatment rather than prevention or accessing dental care.

When full text reading of the thirty-nine Cochrane-sourced articles was completed, sixteen articles were excluded for the following reasons: Not a carer led report (7), Not oral hygiene focused (6), Not a chronic condition (2), Full text not available (1), Non-English language (1). Of the remaining 23 Cochrane sourced articles, only 8 of these articles had met the criteria for inclusion in the Cochrane Review of oral hygiene interventions for people with ID.

A further thirty-eight articles were excluded from the Realist Search as they were not oral hygiene focused articles. The remaining fifty-eight articles were assessed for relevance and rigour and data were extracted using NVivo 11.

5.6 Step 4: Data Synthesis

Characteristics of the studies

This section will present a summary of studies included in the review, the types of carer-led oral hygiene interventions identified and the populations involved.

Of the 58 articles included in this review, 14 specifically involved an oral hygiene intervention for people with ID; three of these articles referred to the same study (Mac Giolla Phadraig *et al* 2013, 2015 and 2016). Of the twelve interventions described, five focused on increasing the oral health knowledge and toothbrushing skills of the carers (Nicolaci & Tesini 1982, Davis & Whittle 1990, Fickert & Ross 2012, Gonzalez *et al* 2013, Mac Giolla Phadraig *et al* 2013, 2015), one of these also assessed the behaviour and attitude of the carers (Mac Giolla Phadraig *et al* 2013, 2015). Two of these studies used a 'train-the-trainer' approach (Nicolaci & Tesini 1982, Mac Giolla Phadraig *et al* 2013, 2015). One intervention used a multi-team approach, involving multiple healthcare professionals and administrative support staff, to implement individualised oral care plans (Altabet *et al* 2003). Two interventions focused on training the carers to coach the people with ID for whom they cared (Kissel *et al* 1983, Glassman & Miller 2006), another focused on the use of rewards by the carers (Willette & Savage 1978), a third used a moral contract approach along with training, re-enforced training and provision of resources (Binkley *et al* 2014) and a fourth used the introduction of an new oral hygiene policy, along with training and monitoring of the carers adherence to the policy (Lange *et al* 2000).

All of these twelve oral hygiene studies involving people with ID had been identified in the Cochrane search, eight of which (67%) had been included in the Cochrane Review, the remainder not meeting the Cochrane review criteria.

There were six articles reporting qualitative surveys of carers of people with ID (Wadsworth *et al* 1986, Cullen-Erickson 1994, Crawford & Drinnan 2007, Zizzi *et al* 2014, Chadwick *et al* 2017, Eijsink *et al* 2018) and one systematic review of the topic (Hithersay *et al* 2014).

Of the studies including people with ID, the majority included people with mixed levels of ID (N=15). Two included people with severe ID only, one with moderate ID only, one with both mild and moderate ID only and one with mild ID only.

The remaining 23 oral hygiene interventions, included in the review, were of populations other than those with ID only. They involved elderly people (Simons *et al* 2000, Wardh *et al* 2003, Nicol *et al* 2005, Gammack & Pulisetty, 2009, Shimoyama & Suzuki 2007, De Visschere *et al* 2011, Duane *et al* 2011, Blinkhorn *et al* 2012, De Visschere *et al* 2012, Van Der Putten *et al* 2013, Janssens *et al* 2016, Kohli *et al* 2017, Janssens *et al* 2018), people with dementia (Wardh *et al* 2002, Kullberg *et al* 2009, Forsell *et al* 2010, Kullberg *et al* 2010, Forsell *et al* 2011, Jablonski-Jaudon *et al* 2016), or people with disabilities (physical, mixed or unspecified) (Adiwoso & Pilot 1999, Faulks & Hennequin 2000, Colodny *et al* 2015, Pradhan *et al* 2016).

Most of the articles reporting interventions were of people in institutional settings (42). Two were based in day centres, two were based in mixed settings, two were based at the individual's home and one was based in an orphanage. The remaining nine articles were review articles, so the settings were mixed.

The review articles and qualitative surveys were on the topic of providing oral hygiene care to others (Brady *et al* 2006, Chiba *et al* 2009, Thole *et al* 2010, Weening-Verbree *et al* 2013, Coker *et al* 2014, De Lugt-Lustig *et al* 2014, Minihan *et al* 2014, Risma *et al* 2015, Barnes *et al* 2016, Hammar *et al* 2016, Wang *et al* 2015, Albrecht *et al* 2016, Goh *et al* 2016, Shah *et al* 2017).

Of the 58 articles, 20 were reporting the quantitative outcomes of an intervention, six were reporting both quantitative and qualitative outcomes, thirteen were reporting qualitative outcomes only, eight were review articles and one was presenting a protocol and qualitative outcomes of a pilot study.

The included studies and the theories they provided data for are listed in Table 64.

Table 64: Realist Review studies & input into the Candidate Theories

AUTHOR ID	YEAR	T1	T2	T3	T4	T5	T6	T7	T8	T9	T10
Adiwoso & Pilot	1999			X	X	X	X		X	X	X
Albrecht <i>et al</i>	2016	X				X			X		
Altabet <i>et al</i>	2003	X				X	X				X
Aylward <i>et al</i>	2003	X		X			X				
Barnes <i>et al</i>	2016	X		X		X	X				
Binkley <i>et al</i>	2014	X	X		X	X	X	X	X	X	X
Blinkhorn <i>et al</i>	2012	X		X	X	X	X	X	X	X	X
Brady <i>et al</i>	2006	X		X		X	X		X	X	X
Chadwick <i>et al</i>	2017	X		X	X	X	X	X	X	X	X
Chiba <i>et al</i>	2009	X		X		X				X	X
Coker <i>et al</i>	2014	X		X		X			X		X
Colodny <i>et al</i>	2015			X		X	X		X		
Crawford <i>et al</i>	2007	X		X	X	X	X	X		X	X
Cullen- Erickson	1994					X		X			
Davies & Whittle	1990			X		X				X	
De Lugt-Lustig <i>et al</i>	2014			X	X	X			X		
De Visschere <i>et al</i>	2011	X			X	X				?	
De Visschere <i>et al</i>	2012	X				X			X		X
Duane <i>et al</i>	2011	X		X	X	X	X	X	X	X	X
Eijsink <i>et al</i>	2018					X					
Faulks & Hennequin	2000	X	X	X	X	X	X	X	X	X	X
Fickert & Ross	2012	X				X		X	X	X	
Forsell <i>et al</i>	2010	X		X	X					X	X
Forsell <i>et al</i>	2011		X		X	X			X	X	
Gammack & Pulisetty	2009	X			X	X					
Glassman & Miller	2006	X		X		X	X	X	?	X	
Goh <i>et al</i>	2016	X		X		X		X			
Gonzalez <i>et al</i>	2013					X					
Hammar & Meranius	2016	X	X		X	X					
Hithersay <i>et al</i>	2014	X		X		X				X	X
Jablonski-Jaudon <i>et al</i>	2016				X	X					
Janssens <i>et al</i>	2016	X		X		X		X	X		X
Janssens <i>et al</i>	2018			X	X		X			X	
Kissel <i>et al</i>	1983	X		X		X	X	X		X	
Kohli <i>et al</i>	2017	X		X	X	X					X
Kullberg <i>et al</i>	2009			X	X	X			X		
Kullberg <i>et al</i>	2010		X						X	X	
Lange <i>et al</i>	2000	X		X			X	X		X	X
Mac Giolla Phadraig <i>et al</i>	2013	X			X	X	X	X	X		
Mac Giolla Phadraig <i>et al</i>	2015										
Mac Giolla Phadraig <i>et al</i>	2016	X					X			X	X
Minihan <i>et al</i>	2014	X				X	X				
Nicol <i>et al</i>	2005	X		X	X	X		X	X	X	X
Nicolaci & Tesini	1982	X	X			X	X	X	X	X	X
Pradhan <i>et al</i>	2016	X		X	X	X	X	X	X	X	
Risma <i>et al</i>	2015					X				X	
Shah <i>et al</i>	2017	X			X	X					

Shimoyama & Suzuki	2007		X				X	X				X
Simons <i>et al</i>	2000		X		X	X	X	X	X	Y	X	
Thole <i>et al</i>	2010				X	X	X				X	
Van Der Putten <i>et al</i>	2013		X	X			X		X	X	X	
Wadsworth <i>et al</i>	1986		X				X					
Wang <i>et al</i>	2015		X		X					X	X	X
Wardh <i>et al</i>	2002				X	X	X	X		Y		
Wardh <i>et al</i>	2003		X		X	X	X	X		Y		X
Weening-Verbree <i>et al</i>	2013					X		X	X	X		
Willette & Savage	1978		X						X	Y		X
Zizzi <i>et al</i>	2014					X	X					

Data Synthesis

This section will present a synthesis of the data for each theory followed by a framework of how the refined theories are expected to impact on oral hygiene outcomes.

Each theory has been given a title which broadly summarises it's content; the synthesis for each theory will be presented separately, in the following way:

- A summary of the Expert Panel's thoughts in relation to the clarity, relevance and applicability of the theory in practice along with any general comments from them.
- An overview of the quantity of data extracted from the articles that were relevant to the theory.
- Presentation and discussion of the patterns identified, within the article's mechanisms, contexts and outcomes of relevance to the theory. Some direct quotations from the articles relating to the patterns will be used as examples.
- A summary of the findings, their impact on the Candidate Theory and any refinements made to the theory based on the data.

After the findings for all the theories have been outlined, a table containing the original Candidate Theories and the Refined Theories will be presented to allow comparison.

1. Organisational involvement

Theory 1: Oral hygiene interventions planned at the systems level, embedding values into management practices, will ensure that oral hygiene practices for people with an intellectual disability become established as norms throughout the organisation, encouraging people at all levels to be accountable.

Although the Expert Panel ranked this theory low in relation to relevance, they clearly felt it was important based on their comments, with one stating it was the most important theory. They recognised the role of the systems-level approach in designing, implementing and sustaining quality oral hygiene care policies and the issues that may facilitate or create barriers with this approach.

Forty articles had data relevant to this theory. All of the articles related to formal carers in institutionalized settings. There was overwhelming agreement in the articles that planning at the systems level was essential, the issues arising from the articles related to methods used or recommended to embed oral hygiene practices into daily routines, what a system wide approach might look like and the barriers to a systems level approach.

As with the Expert Panel, the authors of the articles recognised that the systems level approach must be in place from the outset, that stakeholders must be involved at the design and implementation stages as well as their long-term role in sustaining an intervention. Involvement of the whole team was seen as an opportunity to identify the barriers to implementation and sustainability.

"Successful implementation of training must include organizational and system changes" (Aylward et al 2013)

Some examples given in the articles, to help embed the intervention into common practice included the following:

The provision of regular education and skills training by management: as well as keeping the carer's knowledge and skills updated, this was seen as a means to demonstrate to the carers that management felt the issue was important. It

may also encourage learning and development of staff. However, it was felt important that this training be properly resourced.

"Caregiver managers should be encouraged to improve daily oral care for the dependent elderly, be required to promote educational opportunities for general caregivers and to provide caregivers with the necessary information on oral care" (Chiba et al 2009)

"There is an ongoing drive to support staff through National Vocational Qualifications awards. These factors may help foster an environment that is open and receptive to learning and development, encouraging staff to understand and implement recommendations." (Crawford et al 2007)

The introduction of a policy or procedures document: this allowed everyone at all levels to be aware of what was expected in relation to providing oral hygiene care.

"The Dutch guideline "Oral health care Guideline for Older people in Long-term care Institutions (OGOLI)" was developed" (Van der Putten et al 2013)

Formal monitoring: by creating a formal record of care and/or supervision of carers, the introduction of an intervention into the daily routine is facilitated, outlining the level of care expected and allowing senior staff and management to be kept informed.

"designated nurse being required to sign off when oral hygiene had been completed for each resident" (Blinkhorn et al 2012)

"OHC sheets were to be completed and signed off daily by carers on duty for accountability purposes (raised by stakeholders)." (Pradhan et al 2016)

Make someone responsible: Delegating responsibility to a carer or senior staff member will aid compliance by other carers to the policy and increase the motivation and self-esteem of the responsible person.

"An oral healthcare team was installed and assigned responsibility for the implementation of the guideline OGOLI and the daily oral healthcare protocol derived from the guideline" (Janssens et al 2016)

Creative implementation: involving the carers and generating interest and enthusiasm for the intervention was seen as important, examples of methods used included an award scheme where an element of competition was generated between settings, and gradual introduction of the intervention to avoid overwhelming the carers.

"In order to ensure the maintenance of standards once two years has elapsed, the care homes will be required to renew their award. This two year renewal process will generate information as to the influence the award has, on both the long term maintenance of award standards and oral health within care homes." [sic] (Duane et al 2011)

Health Service or Government funding: creating national policy or regulation and dedicated funding were seen as methods to raise the profile of the care provided and assist in sustainability.

"In the current Swedish public dental health insurance system, introduced in January 1999, it was decided as a common public health strategy that the cost of dental care for community-dwelling elderly and disabled patients, as well as certain other prioritized groups of the Swedish population, would be extensively subsidized by the Swedish County Councils" (Forsell et al 2010)

The issues identified as barriers included lack of funding or low prioritisation of funding for oral hygiene care, insufficient long-term monitoring, supervision or support for carers, both the unrealistic expectations from staff and concern that they may be overburdened and issues involving the identification of the right person for the right training course. Finally, the need for long-term follow-up of systems level interventions was identified; the impact of a systems level approach may take longer to manifest as a measurable change.

Based on the input from the expert panel and the relevant articles some clarifications were made to the theory, but no major change:

Refined Theory 1: **Organisational Involvement**

Oral hygiene interventions planned, designed, implemented and sustained at the systems level that ideally establish policy, provide regulation and funding, and monitor staff at all levels to be accountable (C) will embed values across an organisation (M) thereby ensuring good quality and sustainable oral hygiene practices are maintained for people with an intellectual disability (O).

2. Policy and guidelines

Theory 2: When designing and implementing interventions, a familiarity with local policies and guidelines will avoid conflict or confusion and increase the engagement and confidence of carers, when delivering oral hygiene practices for people with intellectual disability.

The expert panel almost unanimously felt that this theory should be linked to Theory 1 (The Systems Level Approach) and that the key issue was that policy and guidelines should exist.

The lack of data identified in the included articles specific to this theory supports their view. Only 7 articles made any reference to local issues. They identified making sure the intervention was compatible with the organisation. However, the main issue raised in the articles was the carers concern about ethical issues in relation to forcing a person to do something they were resisting. Having access to the local policy would potentially resolve this issue.

"The ethical issues of consent to treatment were also raised, but the carers generally declined responsibility for such decisions" (Faulks & Hennequin 2000)

"It has been shown that concerns about residents' integrity and privacy among nursing staff may lead to reluctance to conduct oral care" (Kullberg et al 2010)

This theory was felt, by the expert panel, to be linked to Theory 1 and on reflection, separating policy and regulation issues from the systems-level approach was not felt to be appropriate. The limited evidence identified in the studies, supporting this thinking. Reference to policy and regulation were added to Theory 1.

3. Make it meaningful

Theory 3: Carers must be provided with training that is meaningful and attractive to them resulting in their positive engagement and increased motivation to initiate and sustain delivery of oral hygiene practices for people with an intellectual disability.

The expert panel felt this theory was important but expressed their concerns in relation to the negative effect of staff turn-over and identified that a systems-level approach would support this theory.

The articles with data relevant to Theory 3 provided care to populations of people with ID and dementia but predominantly to elderly people. The articles were reporting on qualitative surveys of carers or those for whom they cared and on interventions, as well as literature and systematic reviews of the topic.

There were several key topics that emerged from the articles to describe meaningful and attractive training: the training should be flexible, sensitive, respectful and inclusive of the carers, indicate a holistic approach; taking into consideration the carers other duties, how these might facilitate or prevent the provision of oral hygiene care and provide information to increase levels of advocacy and carers interest in providing care for the people for whom they are responsible.

It was a commonly held view that the provision of training should take into consideration the level of education and learning styles of the carer. The content of the training should be developed with the input of carers: what did they want to know about and what terms do they used to describe them. Training should consider the level of disability of the person with ID for whom they are caring, provide examples that make sense to the carer, provide them with the opportunity to share their experiences in providing the care. Evaluation and feedback after training may allow improvements to be made and confirm it is implementable.

"Items of knowledge and skill of oral care, which caregivers thought needed to be provided, were as follows; cleaning of teeth (brushing)

76 (74.5%); and denture care (cleaning), how to insert and remove dentures, and removal of food debris, 58 for each (56.9%).” Chiba et al 2009

“All four trainers indicated on the staff survey that use of videotape feedback (i.e., actually seeing themselves performing) greatly aided their learning of the training system. In addition, the staff reported that the rehearsal components enhanced their understanding and implementation of the training skills.” (Kissel et al 1983)

“It appears likely that the differing results from these programmes may be related in part to their target populations, content, manner of delivery and support.” (Brady et al 2006)

Training must be provided at flexible times and/or be available in a format that can be reviewed as required, for example, training CDs or accessible online. The training should be practical, all necessary equipment and resources should be supplied or easily available.

“While improving the oral health knowledge and awareness of the caregivers is important, more attention should be given to practical aspects of oral care training.” (Goh et al 2016)

The training should contain information that might increase the carer’s advocacy levels; provide the carer with information about the effect of oral diseases on quality of life, providing the carer with an understanding of why the person with ID might behave in a certain way, which might then affect the way the carer felt about the person with ID and increase their interest in providing care.

“In order to provide clinical demonstrations of the relevance of mouth care, local patients with common oral conditions were invited to each session to discuss their oral problems with the course participants.” (Nicol et al 2005)

The articles supported the fact that training should take into consideration the carers emotional wellbeing, provide reassurance in relation to the care they

provide and the effort it takes. Information should be provided in a manner that will not overwhelm or raise anxiety in relation to the carers ability and self-confidence to apply the skills learnt; allow the carer to feel a sense of achievement. The person providing the training should be someone the carer can relate to, respect, feel comfortable and safe with, feel able to approach and discuss problems with, without feeling inadequate. The training should provide the carer with the skills to cope with challenges and find solutions.

"It was also considered important to encourage the nursing staff members to contribute with their own ideas because the objective was to assure them that they were capable of finding solutions to most of the problems encountered in oral care situations." (Kullberg et al2009)

The main behaviour change techniques (BCTs) that may relate to this theory are those that increase feelings of being supported and increase the confidence of the carer; others might play a role in increasing motivation and skills (Table 65).

Table 65: Behaviour change techniques relevant to Theory 3

Group	Behaviour Change Techniques	Mechanism of Action
Goals and Planning	problem solving, review of behaviour goals, commitment.	Goals, belief about capabilities, motivation
Social Support	social support (practical) social support (emotional)	Social influences
Shaping Knowledge	instruction on how to perform the behaviour	Skills
Natural consequences	instruction about health consequences	Knowledge, belief about consequences
Comparison of outcomes	credible source	Attitude towards behaviour
Identify	valued self-identity	Self-image

Although the expert panel felt that the benefits from this theory may be reduced due to staff turn-over rates, the importance of it for non-formal carers and more permanent formal carers as well as the quality of the data identified,

encouraged me to retain it. The data relevant to this theory provided examples of what is required to make the training meaningful and attractive to the carers. In order to facilitate application of this theory in practice it makes sense to add these examples to the theory.

Revised Theory 3: **Make it meaningful**

Training that is flexible, practical, sensitive, respectful and inclusive of carers and takes a holistic approach; providing information on health consequences for people with ID, while taking into consideration carers other duties (C and M), will be meaningful and attractive to them, increase their levels of self-efficacy, advocacy, positive engagement and motivation (M) to attend training and initiate and sustain delivery of oral hygiene practices for people with an intellectual disability (O).

4. Needs of people with ID

Theory 4: Training for carers that takes into consideration the environmental and personal needs of people with intellectual disability will increase the carers' potential to adapt principles to individual needs, thus increasing the likelihood of them initiating and sustaining delivery of oral hygiene practices for people with an intellectual disability.

The expert panel were divided on this theory, some felt that well trained carers would be able to adapt their training to the needs of the different people for whom they were caring, whilst others felt it was important to train the carers with the particular needs of the people they cared for in mind.

Twenty-seven articles provided data that considered both positive and negative contexts and mechanisms relating to the environmental and personal needs of people with ID, which may affect the carers ability and interest in delivering oral hygiene care practices for people with ID. However, these tended to focus on the barriers to providing care and were posited as possible reasons for failure of interventions. There were few examples of positive elements actually at work in the interventions.

Tailoring the intervention to match the specific level of ID of the people being cared for was identified in a number of interventions:

"The level of mental retardation has an effect on oral health and on the difficulties in obtaining plaque control. These factors were taken into account in the set-up of the programme." (Adiwoso & Pilot 1999)

Providing practical solutions to environmental and social issues was also seen as assisting the carer to provide or support the person needing care with their oral hygiene. These solutions were identified over time by carers through observations and interactions with the person requiring care. Other interventions identified some key elements and positive adaptations, particularly in relation to preventing and minimizing care resistant behaviour (CRB):

"A key element is to determine the preferences the resident has for performing oral care, such as products used and time and place for performing care. This information can be obtained from family or individuals who have knowledge of the residents past behavioural patterns. There are also general approaches that can be used on a trial -and error basis." "Establishing rapport over time also helps build relationships between mouth care providers and nursing home residents." (Jablonski-Jaudon et al 2016)

The use of BCTs suitable for people with ID were identified, including monitoring of behaviour, practical social support, information about others approval and social reward:

"However, Shaw and colleagues demonstrated that if these IDD persons are supervised, encouraged, and motivated by caregivers, their oral hygiene can be improved." (Binkley et al 2016)

Much of the data related to the barriers to providing care which included issues such as the inability of the person requiring care to communicate or understand the need for oral hygiene care, swallowing difficulties and care-resistant behaviour. Other issues, such as the person's autonomy to refuse care, as well as carer-specific issues such as attitudes to providing, or perceptions of need for, oral hygiene care, lack of time and resources and lack of policy or processes for staff to follow, were identified.

The limited data identified in the articles relevant to this theory may relate to the focus of this review and the search terms used, carer-led interventions; perhaps a new search focusing on the needs of the people requiring care may produce more relevant articles.

There was limited evidence in relation to this theory, whilst what was identified appears to support the theory, further evidence would strengthen confidence in it. For the moment, this theory is put on hold pending a more detail search of the literature.

5. Caring for carers

Theory 5: The personal needs and environment of carers must be considered when planning their training, providing opportunities for flexibility and increasing the likelihood of successful training and of them initiating and sustaining delivery of oral hygiene practices for people with an intellectual disability.

The expert panel also rated this theory low in relation to relevance, alongside planning interventions at the systems level. Their comments in relation to this theory showed some disagreement. One commented

"Carers are the key. Identifying and working within their abilities and circumstances is essential." EP1

while another said

"this must form part of an overall care package and the carers should engage with this despite their personal views". EP6

There may have been some confusion, on the part of the expert panel, in relation to this theory. Some of the expert panel may have thought it was solely referring to the logistics of training. Other issues raised by the expert panel were the need for resources to support carers and the differing needs of formal and non-formal carers.

As the focus of the search was on carer-led interventions, it is not altogether surprising that this theory had the most data associated with it; 48 of the 58 articles provided data relevant to this theory.

Based on the Expert Panel's comments and my own thoughts in relation to the possibility that the issues might be different for formal and non-formal carers, I started to categorise the data for both types of carers separately. However, it soon became obvious that this was not the case, particularly when comparing non-formal and the lower paid, unqualified formal carers. The same issues were present for both groups.

In relation to considering the carers when providing training, issues such as flexible delivery times, small group numbers, supportive and relaxed environments, training that is tailored to the educational level, learning styles and language of the carers, arose:

"Continuing education initiatives also have to accommodate a range of abilities of staff, different educational levels, and a mix of learning styles" (Aylward et al 2003)

Trainers and even the carers themselves may over-estimate their knowledge and understanding of oral health issues. Assessing the existing knowledge of the carers appears to be important:

"Half of the nursing staff members were satisfied with the results of the provided oral care. This is a somewhat alarming finding, considering that both gingivitis and persistent dental plaque were predominant findings among the residents" (Forsell et al 2011)

"A survey of nurses, director-supervisors, and nursing home health aides who provided home care in Iowa found that many misconceptions existed about what constituted appropriate oral and dental practices for institutionalized older adults." (Thole et al 2010)

The 'train the trainer' approach was used by a number of the studies; monitoring and ensuring the training was rolled out to the carers arose as an issue:

"After the intervention (a pyramid-based educational programme), only half of the participants in the intervention group (48%) reported that they participated in the educational programme." (Janssens et al 2016)

The benefit of involving the carers in the design, implementation and evaluation of training was discussed in some articles. The carers ability to provide solutions to challenges, identify training that was relevant to carers and the priority of practical skills training over knowledge were reported:

"nursing staff identified three main barriers to the implementation of an oral hygiene program for their residents." (Blinkhorn et al 2012)

The importance of acknowledging the emotional effect of caring for people with disabilities, the need for support, encouragement and demonstration of empathy towards carers was common in the articles.

Those providing training, fellow staff members and other professionals were identified as playing a role in providing this support, both during and after training, but were often poor at doing so. The provision of this support was seen to affect the carers levels of self-confidence, self-efficacy, stress, patience and emotional wellbeing.

"Mass and colleagues found that 90% of all resident care is carried out by health care aides who have a high school education or less, receive little more than a minimum wage, obtain limited training, and receive minimal long-term benefits, recognition, or support for their physically and emotionally intensive work" (Albrecht et al 2016)

"All caregivers need support to assist with at-home oral care, but family caregivers may require special measures. They reported feeling less confident than paid caregivers about their skills assisting with toothbrushing and flossing and responding to behavioural challenges and were much less likely to have learned to assist with at-home oral care through group training sessions." (Minihan et al 2014)

From the evidence gathered it appears there is considerable debate between carer groups in relation to resistance to care being a barrier to providing oral hygiene care. Two studies of general nurses caring for elderly people in nursing homes reported this was not a barrier, while others caring for those with ID and elderly people have reported it as a significant barrier. Whether this difference is due to the level of training or qualification of the carer or to the level of resistance of the people for whom care is being provided, is unclear.

"Personal & lifestyle influences on daily oral care - Behaviours that carers find challenging which impede daily oral care (e.g.,

aggressive, self- injurious & social inappropriate behaviours). Others were concerned that the person would become "hysterical" or aggressive if they tried to support them to clean their teeth." (Chadwick et al 2017)

The attitude of the carer, in relation to the importance or value placed on the oral health of the people they care for, or in relation to their dislike of providing oral hygiene care, may impact on the level of care provided. At times, the issue of disliking the task was due to resistance by the person for whom care was being provided and not the task itself.

"Among nurses, nursing assistants and nurse' aides, the main barriers to providing adequate oral care, include unawareness of the importance of oral health, lack of appropriate knowledge and skills to perform oral health care, and low priority given to oral health care" (De Visschere et al 2011)

"This qualitative research revealed the attitude of nurses and nurses' aides as one of the important barriers towards oral hygiene support." (Janssens et al 2016)

"Evidence from this and other recent studies indicates that residents' communication and behavioral problems contribute to care providers' dislike of doing OHC [oral hygiene care] rather than disliking the actual task of OHC" [sic] (Thole et al 2010)

The articles relevant to this theory have provided insightful information in relation to the attitudes and opinions of carers. This type of qualitative data is uncommon in oral health research. It suggests that mechanisms rather than contexts are more important to this theory. To fully capture this, the following amendment is suggested for this theory:

Refined Theory 5: Caring for Carers

Carers personal attitudes, opinions and need for empathy and support (M) as well as their oral health knowledge, educational level and personal and work environments (C) must be considered when planning their training, increasing the likelihood of successful training and of them initiating and sustaining delivery of oral hygiene practices for people with an intellectual disability (O).

6. A pat on the back

Theory 6: Positive feedback from outcome reviews and support from other staff will provide encouragement to, and aid motivation of the carers to continue delivering oral hygiene practices for people with an intellectual disability.

The expert panel recognised the benefits to the carers from positive feedback and they raised the issue of how to deal with negative feedback. They also felt some of the terminology in the theory could be more self-explanatory, for example use of the terms “other staff” and “outcome reviews”: who are the “other staff” being referred to and do “outcome reviews” relate to evidence-based reviews or coal-face practical, person-centered reviews. The ambiguity of the terms is acknowledged, we intended the “other staff” to include all staff in the organisation, including management, senior clinical staff and work colleagues. The “outcome reviews” was intended to refer to review of the outcomes of the intervention implemented in their setting.

Twenty-six articles, twenty interventions, three systematic reviews and three qualitative surveys all provided data in relation to feedback and support for carers as potential facilitators for carers to continue to engage in oral hygiene practices for people requiring care.

Most commonly the use of feedback, whether given during training, during the intervention (process feedback) or post intervention, was used as a motivator for the carers. However, this feedback was often provided by the researchers and stopped once the intervention was completed. Feedback was given in the form of verbal praise or encouragement following observation of carers actions, or by means of formal monitoring of oral hygiene practices, the results of which were shared with the carers and management; an element of competition was introduced in one study by comparing the results between different units with all staff in the organisation.

An example of the opportunities taken to provide feedback was:

"A report is compiled using the clinical and instrumental assessment results and sent to relevant parties. This is fed back verbally to

carers to ensure they understand the recommendations and the SLP [Speech-Language Pathologist] observes the main carers to ensure they are able to follow the dysphagia management recommendations.” (Crawford et al 2007)

The importance of feeling supported to provide care was identified in several studies, as were the barriers to supplying this support:

“Feeling cared for by both tutors and other carers contributed to their being able to make sense of information” “However, it could not bring such a network into being and one of the disappointments voiced by many participants was that there was not enduring contact between those taking part once the course had ended suggesting that information, alone, was not sufficient to ensure on-going good care.” (Barnes et al 2016)

“It was important for the oral care aides to feel reassured that they were performing their duties effectively, especially the assurance that came from the nurses and the dental team.” (Wardh et al 2003)

Some BCTs were identified by the authors, others were implied:

“Our review points at promising behaviour change strategies that are not commonly used in improvement of oral health care. These strategies were used in a small number of studies and yielded positive results. They focus on increasing memory, feedback of clinical outcomes, and mobilizing social norm. Increasing memory of understanding of information is reached by group discussions, answering questions and clarifying information” (Weening-Verbree et al 2013)

The BCTs potentially in use in the interventions considered for this theory are outlined in the table below (Table 66)

Table 66: Behaviour change techniques relevant to Theory 6

Group	Behaviour Change Techniques	Mechanism of Action
Goals and Planning	problem solving, review of behaviour goals,	belief about capabilities, motivation
Feedback and monitoring	feedback on behaviour and outcomes of behaviour, self-monitoring of behaviour and outcomes of behaviour	feedback processes
Social Support	social support (practical) social support (emotional)	social influences, environmental context and resources
Comparison of behaviour	social comparison, information about others' approval	subjective norms, social influences
Antecedents	restructuring the social environment	behavioural cueing

The evidence from the literature supports the theory that feedback and support for carers can provide them with the encouragement to become motivated to provide care, however some clarifications might improve this theory. The feedback does not need to be only positive feedback, although negative feedback should be measured and delivered sensitively. The feedback should not only be provided at the outcomes stage but throughout the intervention and efforts made to ensure this feedback is built into the long-term plan. Support can be provided by oral care professionals (OCP), staff and management, other carers and from self-reflection.

The theory has been revised with these elements in mind:

Refined Theory 6: A pat on the back:

Sensitively delivered feedback (both positive and negative), at all stages of implementation, evaluation and maintenance of oral hygiene care interventions and support from others (C) will provide carers with encouragement, incentivisation and motivation; improving their levels of self-efficacy and belief in their capabilities (M) to continue delivering oral hygiene practices for people with an intellectual disability (O).

7. Achieving goals

Theory 7: Agreed goal setting, with achievable steps, will provide carers with a reminder of their intention and an incentive to undertake sustained delivery of oral hygiene practices for people with an intellectual disability.

The expert panel supported this theory but wondered who would monitor the goals; one suggested that the goals could be included in a formal carers personal development plan, but they also saw benefits for the non-formal carer. Setting timeframes to achieve the goals was viewed as important.

The articles unanimously supported this theory, most of the data extracted related to ways in which goal setting could be incorporated into interventions to improve implementation and outcomes.

Monitoring of carers behaviour was the most common method used to incentivise and remind carers of their intended behaviour:

"The paperwork for undertaking oral hygiene was similar to that for the medication round, with the designated nurse being required to sign off when oral hygiene had been completed for each resident"
(Blinkhorn et al 2012)

Carers were provided with the opportunity to review and self-monitor their behaviour, as well as being monitored by their superiors. This also allowed some feedback and approval from others. The required paperwork provided cues or prompts to perform the behaviour.

The requirement of most institutions to provide continuing professional development and individuals self-interest in learning was used by many of the interventions as an incentive for the carers to engage with the intended care:

"After each training session, all participants were given a certificate of attendance. A certificate was also provided to each of the care establishments in recognition that their staff had received the training." (Nicol et al 2005)

Similarly, awards or rewards with specific goals were used as incentives:

"The Fife Oral Health Care Award was developed within Fife and based on national guidance and existing national award structures. The criteria included a range of indicators including the proportion of care home staff receiving oral health training, the oral hygiene of residents and the use of denture marking." (Duane et al 2011)

Two interventions used a moral contract to outline the agreed goal or intended care, this contract could indicate commitment and be used as a reminder to the carer of their intention to undertake the care:

"A moral "contract" was agreed upon between carer and dentist that if all treatment was undertaken, including scaling, then the carer would help to maintain the resident's oral hygiene status." (Faulks & Hennequin 2000)

"A moral agreement was agreed upon between the dental team and the trainees." (Mac Giolla Phadraig et al 2013)

Having a plan, aligning the intended behaviour with an existing one and identification of other BCTs that may assist the carer make behavioural changes was seen as important:

"We also identified determinants that have not yet been addressed, i.e. action control and maintenance. Action control entails the use of cues or reminders (Abraham and Michie, 2008; De Bruin et al., 2009), which is shown to be an effective strategy in the implementation of several innovations (Grol and Wensing, 2011). Maintenance entails formulating goals for maintenance of behaviour and relapse prevention (Abraham and Michie, 2008; De Bruin et al., 2009). The effect of the use of these strategies to improve oral health care needs to be studied." (Weening-Verbree et al 2013)

The potential BCTs identified in these extracts include the following (Table 67):

Table 67: Behaviour change techniques relevant to Theory 7

Group	Behaviour Change Techniques	Mechanism of Action
Goals and Planning	goal setting (behaviour), action planning, review behaviour goals, behavioural contract, commitment	Goals, attitude towards behaviour, behavioural cueing
Feedback and monitoring	Monitoring of behaviour by others, with or without feedback on behaviour, self-monitoring of behaviour	Feedback processes
Associations	Prompts/cues, cue signalling reward	Behavioural cueing, environmental context
Reward and threat	Material incentive and/or reward, incentive outcome	Reinforcement
Repetition and Substitution	Habit formation	

All the studies supported this theory and no revision was felt necessary.

8. Support from whom?

Theory 8: Support from oral care professionals will provide opportunities to discuss the barriers and identify solutions to providing oral hygiene care for people with an intellectual disability, increasing self-efficacy, self-confidence and long-term engagement by the carers in sustaining delivery of oral hygiene practices for people with an intellectual disability.

The expert panel recognised the value of support but raised concerns regarding the feasibility and even the need for long-term support from oral care professionals (OCPs) at an organizational level. The value of this support on an individual basis for non-formal carers was acknowledged. The issue of funding and valuing OCPs in this role was raised.

Almost half of the articles in this review referred to the role of OCPs in relation to carer led oral hygiene interventions. However, there was inconsistent findings between the articles in relation to the importance of this role: some reported that regular input from OCPs was vital to the success of an intervention, others said that as long as there was regular support, it did not matter who was providing it, some stated that they felt more input from OCPs was required.

An example of a successful oral hygiene interventions that involved OCPs throughout the intervention concluded:

"Conclusions: The oral healthcare programme including a mobile dental team resulted in a significant increase of the care staff knowledge and attitude regarding oral health. Clinical relevance: The integration of a dental professional team in nursing home organisations should be encouraged because it could be valuable to tackle barriers for the provision of daily oral hygiene and to support the continuous integration of oral health care into general care."
(Janssens et al 2018)

The presence of an OCP did not always improve the carer-led oral hygiene, the role carried out by the OCPs could be carried out by other non-OCPs trained in the role:

"The community dental team remained available for support post-training and attended the homes on a regular basis to provide dental care for specific residents. This allowed for even further input and information, but despite this the results still showed no change in health related behaviours of carers." [sic] (Simmons et al 2000)

"The authors encourage other long-term oral hygiene programs using direct care staff. The OTA [occupational therapy assistant] and DCS [direct care staff] were effective oral hygiene teachers because they were most familiar with the behavior and habits of the residents and policies of the institution." [sic] (Nicolaci & Tesini 1982)

Some negativity in relation to presences or absence of OCPs on the team was identified in the interventions and reviews, such as the lack of support, the time requirement and the subsequent demand for dental care for the residents:

"The principal applicant (R.N.), who is a qualified dentist, conducted the training programme with the assistance of a dental hygienist."
"The organization and delivery of the training sessions was time consuming. In addition, there was an increase in requests for professional dental advice and treatment of the elderly residents."
(Nicol et al 2005)

The review articles included in this review reported inconsistent findings:

"A potential explanation for the inconsistent results is the difference in knowledge level, experience and inspiration of the teachers mentioned in these studies: a well-educated health promotor of 15 years' experience [Positive result], a dental hygienist or a nurse educated by a dental hygienist [No change], a non-defined teacher, dental therapists and hygienists with teaching qualification [A positive change in knowledge only], a dental hygienist [no change] and dentists, dental hygienists and competent nurses [A positive

*change in knowledge only].” (De Lugt-Lustig et al 2014) (Content in
[] brackets added by CW for clarification)*

Based on the findings thought to be relevant to this theory, it appears that there is insufficient evidence to support this theory. Some of the context and mechanisms identified in this theory (opportunities to discuss the barriers and identify solutions (C) and increasing self-efficacy, self-confidence and long-term engagement (M)) overlap with other theories – Theory 3; meaningful and attractive training, Theory 6; positive feedback and Theory 9; repeat training. This theory was therefore eliminated.

9. Remind them

Theory 9: Staff turnover and/or rotation as well as fading of knowledge means that knowledge and skills training should be repeated on a regular basis, maintaining levels of engagement, motivation, self-efficacy and self-confidence of carers to sustain delivery of oral hygiene practices for people with an intellectual disability.

The panel of experts acknowledged the challenge of staff turnover and the importance of regular training but were concerned about the feasibility of induction training for all new staff.

The articles relevant to this theory included both reports of interventions, literature reviews and systematic reviews of the topic. Oral hygiene care was reported as being provided to those who required assistance by carers who, for the most part, had minimal healthcare qualifications or were unqualified, with low levels of oral health knowledge or training in providing oral hygiene care. Many of the authors acknowledged the need for repeat training due to the high turnover rate of care staff.

Several authors identified the need for evidence in relation to the role of repeating knowledge and skills training in relation to continuity of oral hygiene care, while others accepted that it played a role:

"Due to sample size it is not possible to know whether this demonstrates a loss of knowledge over time, indicating that oral hygiene knowledge, assessment of skills, and compliance should be revisited on a regular basis." (Fickert & Ross 2012)

Coaching, group discussions and supervision directly after education and training sessions as well as ongoing support and encouragement from oral care professionals (OCPs) were identified as means to motivate and engage the carers long-term.

An example of the possible contexts and mechanisms in action is provided below:

"After school holidays most parents of handicapped children had to employ a new maid or nurse and so a special oral cleaning training exercise was scheduled for the new helper to maintain continuity of plaque removal as required." "OHE was given at the beginning of every school year to the parents to reinforce their knowledge and to increase oral health care for their family members." [sic] (Adiwoso & Pilot 1999).

In the context described in this intervention, new carers were trained as they joined the care team and annual re-training was provided for all carers. The constant support of an oral health educator, by way of regular attendance at the day centre, assisted by the more permanent teaching staff at the day centre who had received training, facilitated the regular reinforcement of knowledge and skills, providing the carers with continual encouragement and support that resulted in continuity of care for the people for whom they were providing care. However, the feasibility of providing this level of support by OCPs long-term is questionable. Possible solutions were identified in other studies:

"They focused on increasing memory through discussions, answering questions and clarifying information... As a consequence, an informal repeated education approach was established" (Janssens et al 2018).

In the intervention quoted above, some existing staff members in the institution were assigned additional roles in relation to oral health: a coordinator, who was usually a nurse or speech and language therapist and one oral care aide per ward, who were usually nurses or nurse aides. These appointments were based on a State-implemented, oral health protocol. Training was received at management and nursing levels in the institution, which was specific to the roles for which they were responsible, with the ultimate aim of having the oral care aides train their nurse's aide colleagues in the provision of the oral hygiene care to their clients using a 'train the trainer' concept. This approach facilitated regular informal exchange of information between peers and provided a constant source of support to the nurse's aides.

This assisted in the retention of knowledge, increased coping skills and thus reduced the barriers to providing consistent oral hygiene care to their clients.

"One potential solution is to bring daily oral hygiene care within the scope of care of qualified nurses and ensure it is core business. The qualified staff turnover in residential homes is less than unqualified staff, so their long-term potential to effect change is possible." "the nurses were in charge of the program and did not delegate it to unqualified staff, many of whom changed jobs frequently." (Blinkhorn et al 2012).

In this intervention, the researcher identified at the outset the issue of staff turnover in the lower paid carers and opted to involve the more permanent, higher qualified nurses. The oral hygiene care protocol and process was designed and implemented by the nurses who identified the barriers to providing the consistent care required. The nurses had ownership of the process and it became embedded in the ward routine.

The three examples of interventions outlined above are very complex and use many techniques to elicit behaviour change. All three elements of Capability, Motivation and Opportunity are present in the interventions. Using the behaviour change wheel (Michie et al 2011), the behaviour change intervention functions identified in these interventions are education, training, enablement and environmental restructuring and possibly the policy categories of regulation and guidelines. Some of the BCTs identified in relation to repeat knowledge and training, which may assist in the replication of the CMOs required, are listed in Table 68.

Table 68: Behaviour change techniques relevant to Theory 9

Group	Behaviour Change Techniques	Mechanism of Action
Goals and Planning	goal setting (behaviour and outcome), problem solving, action planning, review of behaviour goals, commitment.	Goals, belief about capabilities, motivation

Feedback and monitoring	feedback on behaviour	Feedback processes
Social Support	social support (practical) social support (emotional)	Social influences
Shaping Knowledge	instruction on how to perform the behaviour	Skills
Natural consequences	instruction about health consequences	Belief about consequences, knowledge
Comparison of behaviour	social comparison	subjective norms, social influences
Comparison of outcomes	credible source	Attitude towards behaviour
Antecedents	restructuring the social environment	Behavioural cueing
Identify	identification of self as role model valued self-identity	Self-image

The evidence identified in relation to Theory 9 is supportive of the theory. The evidence additionally suggests that to reduce the effect of staff turnover, which occurs most often amongst the lower-qualified staff, the responsibility of providing oral hygiene care, or training others to provide this care, should be given to respected or valued members of staff, who may be the permanent or more highly qualified staff. These staff members may have greater levels of oral care knowledge, autonomy, follow-through and commitment and be better able to communicate with the other staff members, instilling teamwork and advocacy on behalf of the people for whom they care. The theory was refined to include these elements.

Refined Theory 9: Remind them:

Regular re-training of staff who are respected/valued within their workplace and who are given ownership of this training to train other staff (C), will reduce knowledge fading, maintain motivation and increase the coping skills, self-efficacy and self-confidence (M) of all staff to sustain delivery of oral hygiene practices for people with an intellectual disability (O).

10. Everyone's voice

Theory 10: A multi-stakeholder team who have designed, implemented and evaluated an intervention will feel supported and appreciated, will have considered all of the complexities and be organised and prepared to deal with any issues, which will increase the sustainability of the intervention.

The expert panel labelled this theory "In the best of worlds"; they were conscious of the challenges involved in achieving it, but felt it was important. Some confusion was evident in relation to the multi-stakeholder's role in evaluation of an intervention.

A small number of studies reported involving key stakeholders from the outset and providing specific training for each of the stakeholders, dependent on their background and educational level. The advantages of involving all stakeholders were highlighted in a number of studies:

"Carers made suggestions for service improvement such as SLPs [Speech-language pathologists] spending more time getting to know clients outside of mealtimes, providing videos of appropriate feeding support methods for an individual client, and providing a step-by-step laminated list of recommendations for individual clients." "This also dictates the need for multidisciplinary liaison because in the majority of cases the prescription of utensils is done by occupational therapists in the region." "Within this training, staff are encouraged to collaborate on the development of guidelines for individuals." (Crawford et al 2007)

The involvement of specialists was also seen as valuable, and their expertise was sought when needed. However, this tended to be in large hospital or institutionalised settings, where these specialist staff were on site.

"Most of the children had insufficient head control and needed extra care and exercise from the physiotherapist to promote better head stability. Stimulation of the facial muscles was exercised by the speech therapist and occupation therapist, assisting in the tooth

brushing training. Tooth brushing techniques were adjusted according to the neuromuscular conditions.” (Adiwoso & Pilot 1999)

Challenges to the workings of a multi-stakeholder team such as hierarchical issues or differences in education level were identified:

“However, many difficulties face care professionals when attempting to work together as members of a multidisciplinary team because of differences in the levels of education, professional knowledge and experience. There is also a gap between specialist knowledge and practice amongst caregivers.” “It is also important for each related member of the oral care professional body to mutually comprehend the role of other staff.” (Chiba et al 2009)

When this theory was initially developed, the concept of a multi-disciplinary team included as stakeholders, people with ID and their carers. However, for the most part, the included studies described a multi-disciplinary team as consisting of senior clinical staff, medical specialists and administrators. The inclusion of the individuals requiring care or their formal or non-formal carers on the team was rare, as was their involvement in the design or implementation of the intervention. Only one study considered the importance of each team member understanding the role of the others; it did not include the people for whom care was being provided or their carers. Given the acknowledged importance of engaging the carers in oral health interventions, the lack of evidence regarding their presence when designing and implementing interventions is a cause for concern. A more focused review of the literature is required to confirm or reject this theory. For the moment, the theory is put on hold pending a more detailed search of the literature

5.7 Step 5: Refined Theories

5.7.1 Summary of the Mechanisms, Contexts and Outcomes

The Mechanisms

To better describe the mechanisms identified in this Realist review, it is helpful to consider their two constituent elements: resource and reasoning. The resource element tends to be more concrete or visible, described by Dalkin and colleagues (2015) as “the component introduced in a context”, while the reasoning element is invisible and can be ‘triggered’ in an individual along a continuum, resulting in a change in thinking or behaviour that can happen slowly over a period of time, or rapidly like an ‘on/off switch’. Both elements of the mechanism work together.

For example, in an organisation (Context) *monitoring of staff at all levels* (Resource mechanism) results in staff feeling *accountable, incentivised and motivated* (reasoning mechanism) to undertake regular oral hygiene practices (Outcome).

The mechanisms identified related to the individual or the organisation, the same mechanisms were seen to be in action in several different contexts (Table 69 and Table 70).

Table 69: Mechanisms at the individual carer level and linked theories

Theory Title	Resource Mechanism	Reasoning Mechanism
Make it Meaningful	Training that is: flexible practical includes information on health consequences for people with ID acknowledges carers other duties	Meaningful Attractive Engaging Motivational Respectful Inclusive
Caring for carers	Carers work environment Carer personal environment Carers attitude, opinion	Self-confidence Self-efficacy Motivation

A pat on the back	Support from other staff Feedback	Support Encouragement Motivation
Needs of people with ID	Environmental and personal needs of the people with ID	Awareness

Table 70: Mechanisms at the Organisational level and linked theories

Theory Title	Resource Mechanism	Reasoning Mechanism
Organisational Involvement	Oral hygiene intervention planning design, implementation Funding Resources Staff Monitored at all levels Government and Local policies and regulations	Embed values Establish norms Accountability Incentivisation Engagement Motivation Self-confidence Clarity
Remind them	Knowledge and skills training Repeat training Peer training Assign responsibility	Knowledge, Self-efficacy Self-Confidence Motivation Engagement
Achieving Goals	Agreed goal setting Rewards or incentivisation	Intention Goal Incentivisation Motivation Encouragement
Everyone's voice	Multi-stakeholder team input Overall vision	Involved expertise appreciated/ valued organised Supported

The Contexts

The contexts identified also differed for the organisation and the carers (Table 71).

Table 71: Context for Organisations and Carers

Organisational Contexts	Carers Contexts
Provision of training/ resources	Educational level of carer
Level of support for carers	Knowledge level / fading knowledge
Existing policies (government /local)	Skills/ ability to adapt skills
Demands on carers – other duties	Carers attitude to oral health
Monitoring of carers	Differing needs of people with ID
Staff turn-over	Personal contexts
Clear steps/goals	Environmental contexts
Stakeholders involved	

The outcomes

The outcomes reported or referred to in the articles related to the organisation, the carers and the people for whom care was being provided (Table 72).

Table 72: Outcomes reported/referred to in the studies

Organisational Outcomes	Carer Outcomes	People being cared for Outcomes
Good quality and sustainable oral hygiene practices maintained	Carers are physically and emotionally supported	Personal and environmental needs are addressed
Regular training	Oral health knowledge and skills improved	Improved standards of oral hygiene Improved quality of life
Oral hygiene practices embedded into normal routine	Oral hygiene practices embedded into normal routine	Improved frequency of oral hygiene practices Improved oral health
Goals achieved	Goals achieved	Goals achieved

5.7.2 Candidate and Refined Theories following synthesis

Presented in the comparison table below are the original Candidate Theories and the Refined theories following synthesis of the data in the included studies (Table 73). To complete this chapter a Framework is presented depicting how it is anticipated that the Refined Theories may work together to impact on

carer-led oral hygiene interventions for people with an intellectual disability (ID) (Figure 36). A discussion of the Realist Review process and the findings will be presented in the next Chapter (Chapter 6) along with the conclusions drawn.

Table 73: Candidate and Refined Theories based on synthesis

Candidate Theories	Refined Theories
Theory 1: Oral hygiene interventions planned at the systems level, embedding values into management practices, will ensure that oral hygiene practices for people with intellectual disability become established as norms throughout the organisation, encouraging people at all levels to be accountable.	Organisational Involvement Oral hygiene interventions planned, designed, implemented and sustained at the systems level that ideally establish policy, provide regulation and funding, and monitor staff at all levels to be accountable (C) will embed values across an organisation (M) thereby ensuring good quality and sustainable oral hygiene practices are maintained for people with intellectual disability (O).
Theory 2: When designing and implementing interventions, a familiarity with local policies and guidelines will avoid conflict or confusion and increase the engagement and confidence of carers when delivering oral hygiene practices for people with intellectual disability.	Policy and Guidelines Included in Theory 1

Candidate Theories	Refined Theories
Theory 3: Carers must be provided with training that is meaningful and attractive to them resulting in their positive engagement and increased motivation to initiate and sustain delivery of oral hygiene practices for people with intellectual disability.	Make it Meaningful Training that is flexible, practical, sensitive, respectful and inclusive of carers and takes an holistic approach; providing information on health consequences for people with ID, while taking into consideration carers other duties (C & M), will be meaningful and attractive to them, increase their levels of self-efficacy, advocacy, positive engagement and motivation (M) to attend training and initiate and sustain delivery of oral hygiene practices for people with an intellectual disability (O).
Theory 4: Training for carers that takes into consideration the environmental and personal needs of people with intellectual disability will increase the carers' potential to adapt principles to individual needs, thus increasing the likelihood of them initiating and sustaining delivery of oral hygiene practices for people with intellectual disability.	Needs of people with ID Limited evidence, which although appearing to support the theory, is not sufficient to include it in the final list. Further detailed search of the literature is required.
Theory 5: The personal needs and environment of carers must be considered when planning their training, providing opportunities for flexibility and increasing the likelihood of successful training and of them initiating and sustaining delivery of oral hygiene practices for people with intellectual disability.	Caring for Carers Carers personal attitudes, opinions and their need for empathy and support (M) as well as their oral health knowledge, educational level and personal and work environments (C) must be considered when planning their training, increasing the likelihood of successful training and of them initiating and sustaining delivery of oral hygiene practices for people with intellectual disability (O).

Candidate Theories	Refined Theories
Theory 6: Positive feedback from outcome reviews and support from other staff will provide encouragement to and aid motivation in the carers to continue delivering oral hygiene practices for people with intellectual disability.	A pat on the back Sensitively delivered feedback (both positive and negative) at all stages of implementation, evaluation and maintenance of oral hygiene care initiatives and support from others (C) will provide carers with encouragement, incentivisation and motivation; improve their levels of self-efficacy and believe in capabilities (M) to continue delivering oral hygiene practices for people with intellectual disability (O).
Theory 7: Agreed goal setting with achievable steps, will provide carers with a reminder of their intention and an incentive to undertake sustained delivery of oral hygiene practices for people with intellectual disability.	Achieving Goals (no revision was felt necessary) Agreed goal setting with achievable steps(C), will provide carers with a reminder of their intention and an incentive (M) to undertake sustained delivery of oral hygiene practices for people with intellectual disability (O).
Theory 8: Support from oral care professionals will provide opportunities to discuss the barriers and identify solutions to providing oral hygiene care for people with intellectual disability, increasing self-efficacy, self-confidence and long-term engagement by the carers in sustaining delivery of oral hygiene practices for people with intellectual disability.	Support from whom? There is insufficient evidence to support this theory. Some of the context and mechanisms identified (opportunities to discuss the barriers and identify solutions (C), increasing self-efficacy, self-confidence and long-term engagement (M)) overlap with other theories – Theory 3: Make it meaningful, Theory 6: A pat on the back and Theory 9: Remind them. This theory was therefore eliminated.

Candidate Theories	Refined Theories
Theory 9: Staff turnover and/or rotation and fading of knowledge means that knowledge and skills training should be repeated on a regular basis, maintaining levels of engagement, motivation, self-efficacy and self-confidence of carers to sustain delivery of oral hygiene practices for people with intellectual disability.	Remind them Regular re-training of staff who are respected/valued within their workplace and who are given ownership of this training to train other staff (C), will reduce knowledge fading, maintain motivation and increase the coping skills, self-efficacy and self-confidence (M) of all staff to sustain delivery of oral hygiene practices for people with intellectual disability (O).
Theory 10: A multi-stakeholder team who have designed, implemented and evaluated an intervention will feel supported and appreciated, will have considered all of the complexities and be organised and prepared to deal with any issues, which will increase the sustainability of the intervention.	Everyone's Voice Lack of evidence - more focused review of the literature is required to confirm or reject this theory.

5.7.3 Framework for Refined Theories

While synthesising the data from the individual theories, it was apparent that there was much overlap of contexts and mechanisms between the theories; the theories are not intended to work in isolation.

As a means to present the findings visually, a framework diagram was developed, which at first glance demonstrates the complexity, but which it is hoped, will help demonstrate how the contexts and mechanisms are interlinked (Figure 36). The diagram can be explained as follows: The theories are focused around four broad contexts: the carer, the person with ID, the organisation and the training (The blue circle, with the four sections). There are many sub-contexts for each of these broad contexts (The coloured text beside each section of the Blue Circle; colour coded to the theory of relevance). For example, when considering the carer, the sub-contexts of level of education or knowledge and personal and work environments need also to be considered;

for the person with ID, their level of ID and the setting or home environment is important; for the organisation, resources and policies will have an impact and for training, the content and timing is relevant. The broad contexts are also dependent on one and other in relation to the individual theories (Arrows linking the broad contexts and the theories). For example, the organisation may resource the training, the carers may attend it and the people with ID may provide the focus.

Similarly, the resource mechanisms (coloured text beside each section of the Blue Circle) and reasoning mechanisms (pale oval shapes) may span the broad contexts. Some mechanisms may be triggered specifically in one context while some mechanisms may be triggered in a number of contexts. For example, the monitoring of carers provision of oral hygiene care (resource mechanism, may be carried out by both the organisation and the carers. The result of the monitoring may result in the carers feeling more motivated and incentivised to carry out the oral hygiene care (specific to them) and may additionally result in the oral hygiene care becoming embedded in the organisation (both organisation and carers). These are the reasoning mechanism which tend to be invisible, and can occur within multiple contexts, for multiple theories. Only what were regarded as key resource mechanisms, have been identify on the diagram (pale oval shapes), the more individual mechanisms such as self-confidence, self-efficacy, engagement or empathy are not listed but are implied in the framework.

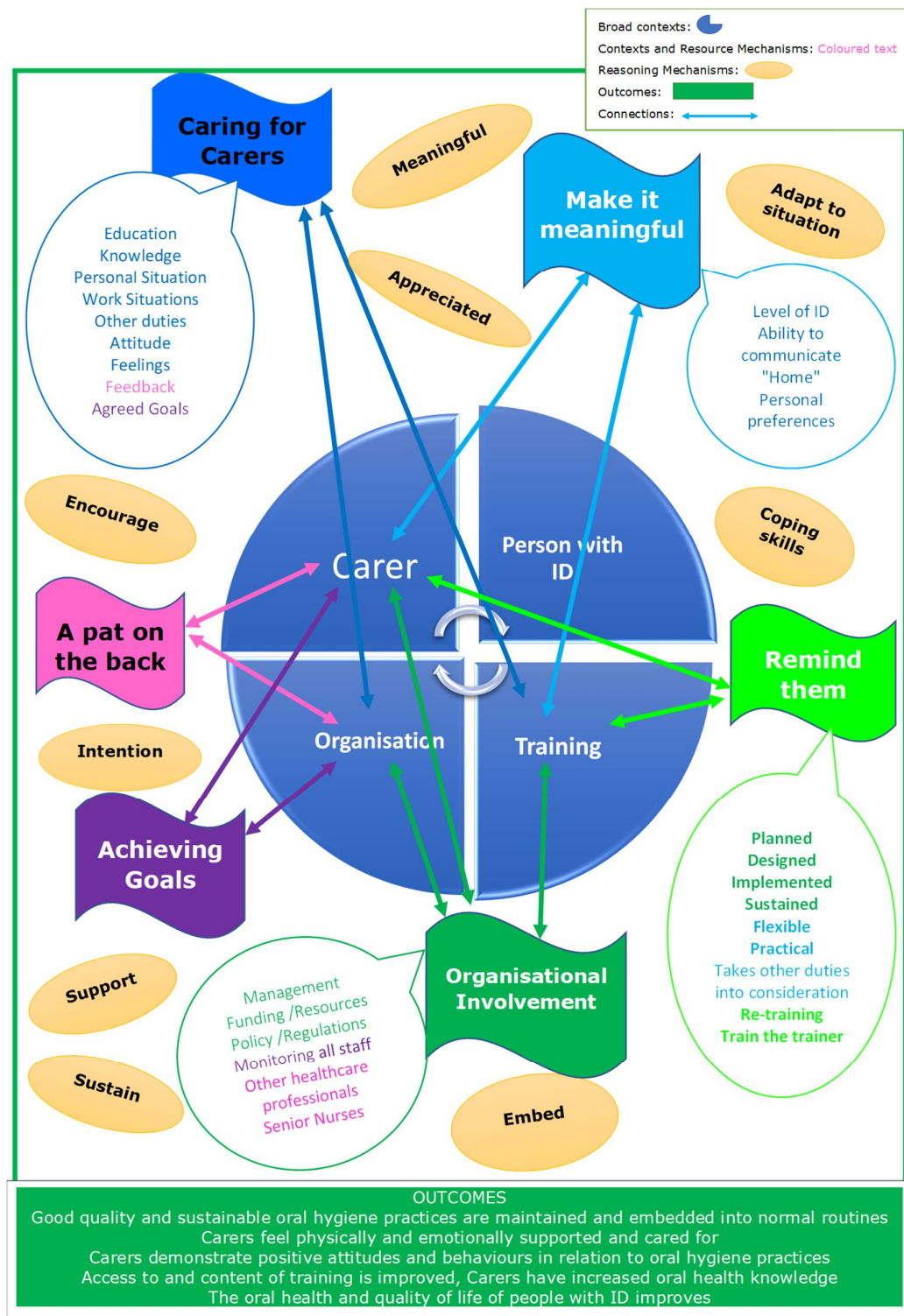


Figure 36: Framework for Theories with key Contexts and Mechanisms

5.8 Discussion

The research question for the Realist Review was “What is it about carer-led oral hygiene interventions for people with intellectual disabilities that works and why?” It was undertaken to identify the contexts and mechanisms that influence the outcomes, implementation and sustainability of carer-led oral hygiene interventions for people with intellectual disabilities.

The use of a realist lens on articles already included in the earlier reviews revealed additional information specific to the research question for this review. The Realist Review also included a broader range of interventions from non-oral healthcare settings and non-ID populations, which provided a rich source of relevant insightful qualitative data.

The Realist Review has allowed the development of a framework which identifies the key contexts, their sub-contexts and the mechanisms that are likely to be triggered for carers of people with ID, the people with ID themselves and the organisations responsible for their care which may impact on the outcomes of oral hygiene interventions for people with ID. As with any Realist Review, the findings are based on the review team’s analysis of the articles/ studies; another team may draw different conclusions. The theories proposed were, from the outset, never stated in order of importance, and this review has not attempted to do this. Nor has it attempted to determine if one theory is reliant on another; although a small number were determined to be either intrinsic to another theory or duplicated by others. What the Realist Review does provide are theories for consideration when planning, designing implementing or sustaining an intervention. A Realist Evaluation of an intervention, which incorporates these theories, is required to assess how these theories impact on the outcomes of a carer-led oral hygiene intervention for people with ID in reality.

There are some limitations to this review: the time frame allowed for this review meant that a follow-up search was not carried out for Theory 4 and Theory 10; relatively small amounts of data were found relevant to both these theories. For theory 4 (The needs of people with ID) this paucity of evidence may have been due to the search terms used, which focused on the needs of

the carer rather than on the needs of the people with ID. Whilst the data that were relevant supported the theory: the value of personal interaction and rapport; respect of autonomy; flexibility and tailoring to the needs of the person with ID, a more detailed search would strengthened confidence in the theory as it stands. For this reason, the theory is excluded from the final list pending a more detailed search of the literature. For Theory 10 (Everyone's voice), there was little evidence that carers, particularly informal carers, and people with ID were considered for inclusion in a team designing and planning oral hygiene interventions for people with ID. There was evidence in relation to the positive role played by other stakeholders. A follow-up search of the non-oral health literature may unearthed evidence to support or reject this theory, in relation to the role of carers and/or the person for whom care was being provided, in the planning of interventions. For this reason, this theory is excluded from the final list until this follow-up search is completed.

Overall, there were minimal changes made to the remaining theories; for the most part changes were made as clarifications rather than changes in thinking. The lack of refinement required may have been due to the fact that this was the third of three reviews on this topic by a team with considerable expertise in the field: the theories developed by a less informed team may have required much greater refinement.

5.9 Conclusions

A Realist Review was undertaken to examine what it is about carer-led oral hygiene interventions for people with intellectual disabilities that works and why. This involved examining the relevant data using a very different lens to that used for the two earlier reviews. Specifically, to identify the contexts and mechanisms that influence the outcomes, implementation and sustainability of carer-led oral hygiene interventions for people with intellectual disabilities.

This was an iterative process that involved much reflection and discussion amongst the review team members. The findings are acknowledged to be subjective rather than objective and influenced by the experiences and personal biases of the team members. What counts in Realist methodology are

the hypotheses that allow us to search for the data and the inferences that are drawn from the data. Theories, with their identified contexts, mechanisms and outcomes, are propositions; are intended to be tested and are open to debate.

The attraction of realist methodology, for me, stems from the practicality of it. The inclusion of common sense in theory development, the resistance to reinventing the wheel, the opportunity to look more closely at what has been done before and to evaluate why and how it worked or not. It strikes me that we have a treasure trove of undiscovered theory in what has been done already, that is being overlooked by the constant pressure to do something new.

The inclusion in this review of studies involving populations other than those with ID, or oral hygiene studies initiated by non-oral health personnel, has emphasised the lack of qualitative data reported in studies initiated by oral health professionals. The value of these data needs to be highlighted; dental schools and centres of research should encourage students and researchers to report qualitative data where it is available, even when the studies are predominately focused on quantitative results.

The implications for future interventions

The refined theories developed from this Realist Review support the belief that the needs of the carers, the content and delivery of their oral care training and support by the organisation, all play important roles in the success of carer-led oral hygiene interventions for people with ID.

Greater time needs to be devoted to the planning and design stages and consideration of which stakeholders should be involved. The need for resources to ensure sustainability, in the form of staff, their training and evaluation, are essential elements of interventions of this type, which must be built into the budget.

To test these theories, a carer-led oral hygiene intervention for people with ID should be designed and evaluated. Consideration of the following elements,

incorporating the refined theories from this Realist Review may assist in this process:

- Stakeholders involvement in the design and implementation of the initiative
- Organisational involvement, providing resources and policy direction as well as continuous support and monitoring of the initiative
- Provision of training that is delivered in a way that suits the carers it is targeting
- Carers personal and environmental needs considered, provision of support for them in their role in the initiative, from all levels of staff
- Consideration of the environmental and personal needs of the person with ID for whom the initiative is aimed
- Goal setting with achievable steps provided to carers
- Regular feedback to carers in relation to the impact of their role on the outcomes of the initiative
- Regular re-training by respected staff members who take ownership of the sustainability of training in relation to the initiative
- Clear statement of how the initiative is expected to work and the behaviour change techniques being used.

Chapter 6 Discussion and Conclusions

6.1 Introduction

The starting point for this thesis was the lack of clear evidence in relation to many aspects of oral health interventions for people with disabilities. Gathering data so that it can be considered and analysed in a systematic way, may provide healthcare professionals with a useful source of reliable evidence to improve the decision-making process in relation to their work practices and thus the health outcomes of the people for whom they care (Abalos *et al* 2001, CRD 2008). The value of both quantitative and qualitative data in this respect is acknowledged. However, when searching and analysing these types of data, different review methodologies are preferable (Kastner *et al* 2012, Tricco *et al* 2016a, Tricco *et al* 2016b).

The aim of the thesis was achieved through the sequential application of multiple methods of data synthesis. Firstly, Scoping Review methodology was applied to systematically search for evidence in relation to what types of oral health interventions have been evaluated for people with disabilities: what types of disabilities have been targeted and what outcomes have been assessed. Secondly, a more focused systematic search for evidence exploring the effectiveness of oral hygiene interventions for people with ID was undertaken following Cochrane Review standards. Thirdly, parallel to the Cochrane Review, Realist Review methodology was used to refine the theories in relation to how carer-led oral hygiene interventions are supposed to work: what are the contexts and mechanisms that may facilitate or create barriers in relation to the success of these interventions.

Chapters 3, 4 and 5 have presented individual discussions of the methodology, methods and findings of each of the three reviews completed for this thesis. In this final chapter, I will provide a brief summary for each of the review methodologies, their key findings and discuss the review specific conclusions. Finally, I will discuss the use of the different methodologies in combination, compare the findings to existing knowledge, and present my overall conclusions and recommendations for future interventions.

6.2 Individual Review Discussion

6.2.1 The Scoping Review

The main purpose of a Scoping Review is to quantify the elements of the existing studies on a broad research topic, such as the populations included, types of interventions undertaken and outcomes assessed. A Scoping Review does not have a defined research question, so keeps the search terms broad and includes all study designs. There is no assessment of the quality or outcomes of the studies and there is no intention to draw conclusions (Arksey & O'Malley 2005, Anderson *et al* 2008).

The Scoping Review, completed as part of this thesis, was a very useful starting point to review an area previously unresearched: oral health interventions for children and adolescents with disabilities. It revealed an increasing interest in researching the outcomes of interventions aimed at children and adolescents with disabilities, 54% of the 112 included studies were reported since the year 2000.

Additionally, it highlighted the existing focus on therapeutic interventions for populations with disabilities (46%). The range of therapeutic interventions reported was very diverse, as was the types of disabilities, resulting in relatively small numbers of studies devoted to each type of therapeutic intervention that focussed on a population with a specific disability.

The next largest group of interventions was skill-based interventions (28%) of which most were focused on oral hygiene skills or methods to teach these skills (90%), and most for a population with ID (48%): this determined the focus of the Cochrane Review in this thesis, which was its main purpose.

The Scoping Review has additionally identified other potential topics for systematic review, for a population with ID. A large proportion of the therapeutic interventions were focused on the prevention of oral diseases (42%). A systematic review would strengthen the evidence in relation to the effectiveness of these types of interventions. The Scoping Review, which included children and adolescents only, identified only a small number of

studies for each type of intervention and did not examine their study design or quality. However, it is likely that individual systematic reviews, if expanded to include all ages groups with ID, on the topics of Chlorohexidine, fissure sealants and fluoride therapies, would have sufficient studies to make comparisons possible and may provide stronger evidence in relation to the effectiveness of these particular interventions.

Finally, this Scoping Review has emphasised an important distinction between therapeutic and skill-based interventions, even when both are focused on preventing oral diseases: skill-based interventions are dependent on the consistent behaviour of the participants. Research by Michie and colleagues (2004, 2008), in relation to the determinants of behaviour and the variety of behavioural change techniques (BCTs) available to influence behavioural change, have demonstrated the complexity of this topic and provided a taxonomy to aid in the identification of the BCTs in action within studies. Given the important impact of behaviour on oral health, future research must give this aspect of oral health interventions greater consideration (Schou 1998, Renz *et al* 2007).

6.2.2 The Cochrane Review

A Cochrane Review, regarded as the gold standard of systematic reviews for information on the effectiveness of health care interventions, looks much more closely at the elements overlooked in a Scoping Review (Agnihotry *et al* 2016). It has a specific research question, with pre-determined criteria for inclusion and exclusion of studies. It gathers all the available evidence, following a systematic approach, thus reducing bias, and results in findings that are more reliable than the individual studies on their own (Higgins & Green 2011).

The objective of the Cochrane Review completed for this thesis was to assess the effects (benefits and harms) of oral hygiene interventions for people with intellectual disabilities (ID). It was an arduous and rigorous process. The title submission, protocol and final review were all reviewed by both the Cochrane Oral Health Group Editorial Team and peer reviewers before each element

could be approved and published. The final draft of the Review is currently with the COHG pending publication, expected in early 2019.

At the outset, there was some concern that a Cochrane Review was not a suitable approach to review interventions for a population with ID given the inherent complexities. This concern was the main reason for the decision to include a wider range of study designs. While such inclusion has resulted in the findings being reported in different formats (narrative and meta-analyses), it has resulted in a very comprehensive review of the types of oral hygiene interventions undertaken for people with intellectual disabilities and the outcomes assessed.

The largest number of studies included in the Cochrane Review examined the effects of using different types of toothbrushes (47%). These studies showed some benefits when a carer of a person with ID used a special manual toothbrush, over a period of four months, and inconsistent findings between outcomes when an electric toothbrush was used by or for people with ID, over periods of between one week and sixteen months. The role of the carer in these studies, either supporting, assisting or carrying out the toothbrushing for the person with ID, was a crucial element.

Training of carers was also examined in the Cochrane Review. The studies included in this comparison (20%), showed that carers oral health knowledge was consistently improved following training, this improvement was shown to be retained for periods between 90 minutes and eleven months. However, training of carers did not always impact on the carers' attitude or behaviour and showed only small reductions in the levels of gingival inflammation of the people with ID for whom they cared. The oral health benefit, to the people with ID, was shown to be greater when some level of monitoring of the carer's behaviour was undertaken. The need for regular provision of the training, both for new carers and to refresh the knowledge and skills of existing carers, was also identified as having an impact on the long-term oral health outcomes.

Importantly, the Cochrane Review also confirmed that people with ID can acquire the skills to undertake some or all of the steps involved in

toothbrushing. Almost a fifth of the included studies examined the effects of teaching people with ID to brush their teeth. The strength of the evidence was low or very low, due to the lack of details reported in the studies or because of the study design used, however all of the studies showed benefits to training people with ID in oral hygiene skills. Four studies assessed changes in behaviour and showed that the toothbrushing skills of the participants improved, in relation to either the number of steps achieved in a toothbrushing routine or an increase in their overall frequency of completing the task of toothbrushing, the remaining two studies also assessed clinical outcomes and these showed that the training impacted positively on the participants oral health.

It became clear in this Cochrane Review, that the level of care provided by carers, in both institutional and home-based settings, to support, assist or carry out the oral hygiene for people with ID and the amount of time given by carers to teach people with ID the skills to brush their own teeth, had a strong impact on the oral health outcomes of the interventions. The level of support provided by management and other healthcare professionals and the level of monitoring of the care provided was also seen to impact on the level of care provided to the people with ID by their carers.

It is also clear from this review, that the standard of reporting in the original research studies, overall, was poor, resulting in many inconclusive findings and low levels of certainty in the evidence. Additionally, the range of outcomes reported was predominately limited to clinical outcomes, specifically, gingival health. There was a noticeable absence of quality of life, patient-centred outcomes. This will be discussed in more detail further on in this chapter.

6.2.3 The Realist Review

The final review carried out as part of this thesis was a Realist Review. The purpose of a Realist Review is to look at how, why, when and for whom comparable interventions work, rather than if they work. It does this by looking closely at the contexts and mechanisms within the interventions to see if there are any patterns which might impact on the outcomes (Pawson 2006).

Whilst it has some methodological elements that are similar to the two earlier reviews, particularly in relation to the search strategy and management of the data, a Realist Review also has many different elements. The process of developing and refining theories in relation to how, why, when and for whom interventions work, required immersion in the subject area, discussion and brainstorming with subject and methodology experts and review of the data using a very different lens (Rycroft-Malone *et al* 2012, Wong *et al* 2013).

Based on the findings in relation to the role played by carers in the Scoping and Cochrane reviews, the interventions of interest in the Realist Review carried out for this thesis were carer-led oral hygiene interventions, which were defined as: any carer-led intervention that may impact on the duration, frequency and/or effectiveness of oral hygiene practices for people with intellectual disabilities or their carers.

The ten original candidate theories about how, when, why and for whom carer-led oral hygiene interventions for people with ID, work, or don't work, were tested against the literature and refined down to six final theories. The eight theories can be summarised as follows:

- The needs of the carers, the content and delivery of their oral care training and support by the organisation, all play important roles in the success of carer-led oral hygiene interventions for people with ID.
- Greater time needs to be devoted to the planning and design stages of interventions and should include consideration of which stakeholders to involve in this process.
- The need for resources to ensure sustainability, in the form of staff and their training, and evaluation of the intervention, are essential elements of interventions of this type, which must be built into the budget.

The Realist Review has produced data that are very different to the predominately quantitative data of the earlier reviews. It provided a valuable insight into the contexts and mechanisms that effect carers attitude and behaviour in relation to providing consistent support, assistance or direct care, to people with disabilities.

The physical and emotional toll as well as the ethical dilemmas, of providing this consistent level of care, to people with disabilities, who may often not understand the need for the care and resist it, was startling. The need to show understanding and empathy and provide support to the carers, to tailor their training to meet their personal and workplace needs, and to monitor the care they provide to the people they are caring for, were all identified as being important elements to ensure that carers felt supported, took ownership of the care and delivered it consistently.

In addition to the findings in relation to people with ID, the refined theories developed from this Realist Review, in relation to the contexts and mechanisms that may influence the outcomes of carer-led oral hygiene interventions, included oral hygiene studies of populations other than those with ID; populations with long-term health conditions requiring assistance with their oral hygiene care. The refined theories could additionally, be applied to interventions involving the carers of those populations, which included elderly people, people with dementia and people with physical conditions limiting their level of self-care.

6.3 Overall Discussion

The findings, from the three reviews completed for this thesis, have highlighted the value of secondary data. The benefit of systematically gathering and combining quantitative data from similar studies to strengthen the evidence is acknowledged. Despite the fact that much of the qualitative data retrieved from the interventions in the reviews were not formally reported as outcomes in the published reports, valuable qualitative findings were also uncovered. The cost of undertaking primary research, as well as the time and effort required from the researchers and participants, makes it imperative that the outputs from the original research are maximised.

Across the three reviews, a number of recurring themes emerged. Three of these themes are explored in detail below, before considering limitations not already considered for each of the individual reviews.

6.3.1 The involvement of carers

The level of involvement of carers in oral health interventions for people with disabilities first appeared in the Scoping Review. However, the details of their role and how their involvement impacted on the outcomes of interventions became more evident in the Cochrane Review. For example, the level of support and assistance provided by them to people with ID in relation to toothbrushing or the impact their oral health knowledge or attitude had on the level of care they provided. The Realist Review further revealed the contexts and resource mechanisms that might support carers to play positive roles more consistently. For example, by acknowledging their input, by providing the practical resources needed, such as time and equipment, and by providing them with regular skills training to carry out their roles.

This builds on primary research that has explored carer perspectives on providing oral hygiene care (Forsell *et al* 2010, de Visschere *et al* 2011, Chadwick *et al* 2013, Johnson 2013, Minihan *et al* 2014, Eijsink *et al* 2018). These three reviews together suggest any intervention involving carers, that neglects to consider the carers' opinions, attitudes, needs, education and personal and workplace environments, is likely to fail.

6.3.2 Stakeholder involvement

The involvement of carers in the design and implementation of general health interventions, for people with ID was shown, in a small number of the studies included in the Realist review, to improve the focus of the intervention and the implementation and sustainability of the intervention, allowing the identification of issues of concern to the carers as well as identification of practical facilitators and solutions to barriers, that are often only obvious to them at the 'coalface', if and when they occurred (Crawford 2007, Blinkhorn *et al* 2011). However, the majority of the oral hygiene interventions that were examined in detail in all three reviews, consulted neither the carers or the people for whom the intervention was targeted, in relation to the design or implementation of the intervention.

Similar conclusions in relation to the importance but lack of involvement of carers, in the design and implementation of oral healthcare interventions, were

found in the other studies of non-ID populations requiring assistance with oral hygiene included in the Realist Review and in a number of systematic reviews of these types of interventions (Brady 2006, Weening-Verbree 2013, Coker 2014). The involvement of the people requiring care, in the intervention design and implementation, was considered to a lesser extent in these reviews, however, it was generally concluded their input would be beneficial.

6.3.3 Outcomes measured

The questions posed in this dental research have, in the past, focused predominantly on clinical and quantitative type outcomes. Thankfully, inclusion of qualitative outcomes in more recent dental research has allowed the profession to gain an appreciation of the value of qualitative data (Stewart *et al* 2008, Petticrew *et al* 2015). Additionally, asking questions about quality of life issues, such as the feelings or opinions of a person with ID or their carers, has increased our understanding of how their life experiences and environment may impact on their oral health behaviour; how they understand or view their oral health (Stewart *et al* 2008).

However, exactly how to assess the qualitative elements, measure change over time and how to connect these changes to our more traditional understanding of what constitutes oral health is complex. In failing to consider the outcomes of importance to people with ID and their carers, or accurately assess changes in behaviour, it may be that valuable benefits have gone unnoticed or unreported in past interventions. This is particularly likely if an intervention has effects across a range of domains, for example in this context, clinical, behavioural and educational domains (Craig 2008).

The research currently being undertaken by Michie and colleagues (2004, 2008) on the use of BCTs may broaden the criteria used to measure success in relation to behavioural change. For example, an intervention that shows an improvement in an individual's self-efficacy can and should be regarded as success.

Additionally, the duration of follow-up may impact on the outcomes observed, or distal outcomes not considered or assessed, may occur. For example,

changes in behaviour noted short-term may not persist long-term or improvements in oral health care may impact on quality of life and have social and economic benefits (Craig 2008).

6.3.4 Limitations

The limitations to each of the individual reviews have been considered in their respective chapters specifically, in the Scoping Review: limiting the participants to children and adolescents and the lack of consideration of the quality and study designs used, in the Cochrane Review; the relevance of findings of statistical significance to clinical significance and the possibility of publication bias, and in the Realist Review; the time available and focus of the review. However, an important element, particular to the Cochrane and Realist Reviews, is worth considering further here: the definition of intellectual disability used. The decision to use the ICD-10 definition in the Cochrane Review was taken with the likely description of ID used in the past studies of interest, in mind. However, the ICF definition, is more holistic and preferable when considering an individual's personal experience of life.

While reviewing the literature around this thesis, it has become obvious that a variety of definitions are still being used in the current literature. Populations that have been excluded from this Cochrane Review of people with ID, have been included in other recent studies and systematic reviews on the same population: people with Autism Spectrum Disorder (ASD) and people with cerebral palsy are two notable groups (Zhou 2018).

The ICD-10 places ASD within the "Disorders of psychological development" section, separate to the section on "Mental Retardation", however, both are positioned under the heading of "Mental and behavioural disorders". Cerebral Palsy is positioned under "Diseases of the Nervous System".

The Diagnostic and Statistical Manual (DSM-5), used by Psychologist to aid in diagnosis, treatment planning and insurance coverage, classifies ASD as a developmental disorder, with or without intellectual disability (ID); some people with ASD may have an above average IQ, others may have below

average IQ (American Psychiatric Association 2013). An American survey in 2014 reported that of the children with ASD who had IQ scores available, nearly a third also had an intellectual disability (Baio *et al* 2018).

A systematic review of the rates of co-occurring impairments, diseases, and functional limitations for people with cerebral palsy (CP) reported that one in two people with CP had an intellectual disability (48%) and more than one in five had a severe intellectual disability (28%) (Novak 2012).

The issue with these populations is that only some people with these conditions would come under the ICD-10 definition used in this thesis. Including people with ASD in a systematic review, for example, some of whom have an IQ of above 70, would confound the outcomes of the review, as their cognitive and physical abilities could be considered to be similar to a general population.

Within the Cochrane Review completed in this thesis, studies that identified that they were focused specifically on populations, of which a proportion may have ID, such as ASD and Cerebral Palsy, or did not clearly state that there was a population with ID reported separately in the study, were excluded from the review.

This may be considered as a limitation, which might have impacted on the findings. However, until such time as the ICF classification is more widely used in research, with agreed population 'core sets' making the process a more practical one, allowing mapping of the ICF classification onto the existing descriptions of impairments, I believe it is, for the moment, the more accurate way of assessing the impact of the interventions on a population with ID. Development of ICF core sets describing disability and oral health are being investigated by different groups, it would be important that an agreed core set is used in future research (Faulks *et al* 2013, Fujita *et al* 2015 (Japanese), Vale *et al* 2017, Dougall *et al* 2018).

6.3.5 Additions to existing knowledge

The Scoping Review is believed to be the only systematic review of the types of oral health interventions targeting children and adolescents with disabilities

in existence. As such, it has provided new knowledge in relation to the population groups targeted and diversity of oral health interventions undertaken. This information is useful to guide the focus of future interventions and to assist in determining suitable topics and populations for systematic review, in relation to having sufficient but manageable numbers of studies to review.

The Cochrane Review is the first oral health Cochrane Review undertaken on a population with intellectual disabilities. It has pushed the boundaries in relation to the conventional study designs included and thus the tools used for the assessment of the risk of bias in the studies, in the Cochrane Oral Health Group. Aside from the findings from the analyses of the combined studies, this review has provided new insight in relation to the types of study designs used, the outcomes assessed and the quality of the studies undertaken for this population. This new information provides reassurance that RCTs are possible for the study of populations with ID and are the preferred study design if the certainty of the clinical evidence is to improve. It also identifies some outcomes that are overlooked, such as quality of life and dental caries, and the need for studies of longer duration.

In relation to new evidence, prior to the commencement of this Cochrane Review, no systematically reviewed evidence existed for this population. However, there have been two non-Cochrane systematic reviews of oral health interventions for people with ID, both published in the last year (Bonardi *et al*, April 2018, Zhou *et al*, December 2018), with which some comparisons can be made.

The Bonardi *et al* review (2018) in the US focused on four areas of oral health: education and behaviour, access to oral health services, prevention of oral health problems and use of sedation. The term 'Intellectual and Developmental Disability' (IDD) was used to describe the population. However, no exact definition was provided. The search terms used in relation to the population included the terms cerebral palsy, physical disability and neurodevelopmental disorder, as well IDD and mental retardation; this included studies of populations with autism. They searched a wide selection of databases and grey

literature but limited the search to English language studies between 1990 and 2013 (Bonardi *et al* 2017). The findings from the studies included under education and behaviour can be compared to this Cochrane Review.

The published document drew conclusions in the form of 'evidence based and promising practices' statements, however, no data or details of the analyses were provided.

The Zhou *et al* Review (2018) focused on interventions that prevent dental caries and periodontal diseases and restricted the review to children and adolescents with ID. The authors searched four databases from the date of their commencement to April 2017, the search was limited to English language studies and included only RCTs or controlled trials. Studies were included in the review utilising a variety of terms used to describe populations with ID, regardless of the level, and included populations with autism. The findings from interventions that examined the effectiveness of oral health promotion activities and the mechanical removal of plaque in the Zhou *et al* Review (2018) can be compared to the findings of this Cochrane Review.

Table 74 provides a summary of the findings of this Cochrane Review compared to the findings of the other systematic reviews, where comparisons can be made. As no details of the data from the Bonardi *et al* (2018) Review are available it difficult to identify the possible cause of the different finding for the use of electric toothbrushes, other than to note that populations with other developmental conditions and a wider range of study designs were included in this review. Additionally, the findings from this review included grey literature and non-peer reviewed studies that were "considered effective by clinicians and their patients, but successful oral health outcomes have not yet been demonstrated with robust research methods". Apart from some inconsistency in the findings for electric toothbrushes, the conclusions for the other comparisons in the three reviews (Special manual toothbrushes, training of carers and training of people with ID) were generally the same.

Table 74: Findings compared to other systematic reviews

(Benefits found = **+**, No benefits found = **-**, No relevant studies = **0**)

Comparison	Cochrane Review	Bonardi <i>et al</i> 2018	Zhou <i>et al</i> 2018 (≤18yrs)
Special Manual TB	+	+	0
Electric TB	-	+	-
Training of Carers	+	+	0
Training of People with ID	+	0	0

The number of studies included in each comparison in the Cochrane Review is larger, having either searched more databases or with a wider timeframe and included non-English studies, as well as only including studies that either had a control group or had multiple measures pre and post intervention, all of which strengthens the evidence.

This Realist Review is believed to be the first qualitative review of carer-led oral health interventions for people with ID. In support of the methodology used, the Realist Review can be compared to a systematic review of carer-led general health interventions to monitor, promote or improve the physical or mental health of populations requiring assistance with their general health, in a variety of settings, which was carried out with an ID population in mind (Hithersay *et al* 2014). Of the 24 studies included in the Hithersay review, few were specifically targeted at carers of people with ID, and of the 12 studies that examined health promotional interventions, none specifically targeted people with ID (Hithersay *et al* 2014). Both this Realist Review and the Hithersay review have highlighted the paucity of qualitative research for people with ID or their carers. Looking to other populations who require care for chronic conditions appears to be a useful source of comparable qualitative issues. Although examining a different element of health, and not using Realist Methodology, similar conclusions were made in the Hithersay review (2014) in relation to the issues of high turnover of carers, engaging the carers in the intervention and focusing on practical skills.

6.3.6 Recommendations

In summary, with the findings of all three systematic reviews in mind, the following proposals are made to influence the design and outcomes of future oral health initiatives for people with intellectual disabilities (ID):

- Interventions should focus on preventing oral disease, either with therapeutic or skills-based interventions, that are effective, with a strong evidence base.
- When planning an intervention, serious thought and time should be spent at the design stage, involving representatives from all the stakeholder groups, in relation to the objectives, programme theory, implementation, outcomes, duration, evaluation and sustainability of the intervention. In particular, it is essential to ascertain what issues people with ID and their carers feel are important to address and to get input from these specific stakeholders in relation to the possible facilitators and barriers to the success of an intervention.
- The opinions, choices, living environment, resources and ability of the person with ID must always be taken into consideration.
- The welfare of carers, their opinions, need for physical and emotional support, personal and work environments as well as their level of education and learning styles, must always be incorporated into the planning and implementation.
- Long-term support, to ensure sustainability, is essential:
 - From Government Health Departments in the form of fiscal resources, health legislation and policy direction
 - From local Management in relation to the implementation of policy guidelines, in the provision of resources and regular and flexible training for new and existing staff, in goal setting, monitoring of the care being undertaken by staff, and
 - From all healthcare staff in the form of attendance at training, meeting goals for provision of care and encouragement and support of colleagues.
- Validated behavioural change techniques should be incorporated into the intervention if it involves changing the behaviour of any of the participants.

- Quality of life changes or long-term, patient-centred outcomes should be assessed where possible.
- Assessment and reporting of qualitative outcomes should be included in the intervention even when the focus of the intervention is predominately on quantitative outcomes.
- Studies should plan to use the 'core outcome sets' currently being developed by Lamont, Schwendicke and colleagues for periodontal disease and dental caries, to allow greater comparison between studies
- Studies should follow recognised standards for implementing and reporting trials
- Studies should use Randomised Controlled Trial study design where possible.

6.4 Conclusions

The three reviews together have provided a comprehensive insight into oral hygiene interventions for people with ID, which did not exist previously: the types of interventions and populations targeted; the clinical and behavioural outcomes; and some contexts and mechanisms that need to be considered to ensure success in future interventions. Each review has provided information that would not have been discovered by the others alone.

Reviewing evidence using parallel approaches has provided depth to the findings reported in this thesis. All of the review approaches have their strengths and limitations: using them in combination, I believe, has resulted in a whole that is greater than the sum of their parts.

This thesis has identified, appraised and synthesised the evidence regarding oral hygiene interventions for people with intellectual disabilities, and provides both practical and theoretical guidance for future interventions.

Chapter 7 References

7.1 General References

Abalos, E., Carroli, G., Mackey, M. & Bergel, E. 2001. Critical appraisal of systematic reviews. *In: The World Health Organization (ed.) The WHO Reproductive Health Library*. Geneva.

- Abraham, C. & Michie, S. 2008. A Taxonomy of Behavior Change Techniques Used in Interventions. *Health Psychology*, 27, 379-387.
- Agnihotry, A., Fedorowicz, Z., Worthington, H. V., Manheimer, E. & Stevenson, R. G. 2016. Systematic reviews in oral health: a quality imperative. *Journal of Evidence-Based Medicine*, 9, 47-52.
- Ainamo, J. & Bay, I. 1975. Problems and proposals for recording gingivitis and plaque. *International Dental Journal*, 25, 229-135.
- Ajzen, I. 1991. The theory of planned behavior. *Organizational Behavior and Human Decision Processes*, 50, 179-211.
- Ajzen, I. & Fishbein, M. 1980. Understanding attitudes and predicting social behaviour. Englewood Cliffs, N.J., Prentice-Hall.
- Akgun, E., Yoldas O & Dogan, C. 2012. Oral health status of children with mental disability: two-year follow-up. *International Association of Disability and Oral Health*. Melbourne: Journal of Disability and Oral Health.
- Al Kindi, N. A. & Nunn, J. 2016. The use of the BDA Case Mix Model to assess the need for referral of patients to specialist dental services. *British Dental Journal*, 220, 401-406.
- Albrecht, M., Kupfer, R., Reissmann, D. R., Mühlhauser, I. & Köpke, S. 2016. Oral health educational interventions for nursing home staff and residents. *Cochrane Database of Systematic Reviews*.
- American Psychiatric Association 2013. Diagnostic and statistical manual of mental disorders. 5th ed. Washington, DC: American Psychiatric Association
- Anders, P. L. & Davis, E. L. 2010. Oral health of patients with intellectual disabilities: A systematic review. *Special Care in Dentistry*, 30, 110-117.
- Anderson, S., Allen, P., Peckham, S. & Goodwin, N. 2008. Asking the right questions: scoping studies in the commissioning of research on the organisation and delivery of health services. *Health Research Policy and Systems*, 6.
- Arksey, H. & O'Malley, L. 2005. Scoping studies: towards a methodological framework. *International Journal of Social Research Methodology*, 8, 19-32.
- Asimakopoulou, K. & Newton, J. T. 2015. The contributions of behaviour change science towards dental public health practice: a new paradigm. *Community Dentistry and Oral Epidemiology*, 43, 2-8.
- Axelsson, P. & Lindhe, J. 1978. Effect of controlled oral hygiene procedures on caries and periodontal disease in adults. *Journal of Clinical Periodontology*, 5, 133-51.
- Axelsson, P., Nystrom, B. & Lindhe, J. 2004. The long-term effect of a plaque control program on tooth mortality, caries and periodontal disease in adults. Results after 30 years of maintenance. *Journal of Clinical Periodontology*, 31, 749-57.
- Aylward, S., Stolee, P., Keat, N. & Johncox, V. 2003. Effectiveness of Continuing Education in Long-Term Care: A Literature Review. *The Gerontologist*, 43, 259-271.
- Baio, J., Wiggins, L, Christensen, D. I. et al. 2018. Prevalence of Autism Spectrum Disorder Among Children Aged 8 Years — Autism and Developmental Disabilities Monitoring Network 11 Sites, United States, 2014. MMWR Surveillance Summary.
- Bandura, A. & Cervone, D. 1983. Self-evaluative and self-efficacy mechanisms governing the motivational effects of goal systems. *Journal of personality and social psychology*, 45, 1017.
- Bartolucci, A.A. & Hillegass W.B. 2010. Overview, Strengths, and Limitations of Systematic Reviews and Meta-Analyses. In: Chiappelli, F., Caldeira Brant, X. M., Neagos, N., Oluwadara, O. O. & Ramchandani, M. H. (eds.) *Evidence-Based Practice: Toward Optimizing Clinical Outcomes*. Springer-Verlag Berlin Heidelberg: Springer.
- Beck, S. & Yasko, J. 1993. *Guidelines for oral care.*, Cary, IL, Sage Products.
- Bender, R., Bunce., Clarke, M, Gates, S, Lange, S. & Pace, N. I. et al. 2008. Attention should be given to multiplicity issues in systematic reviews. *Journal of Clinical Epidemiology* 61, 857-65.

- Bentley, C. D. & Disney, J. A. 1995. A comparison of partial and full mouth scoring of plaque and gingivitis in oral hygiene studies. *Journal of Clinical Periodontology*, 22, 131-135.
- Binkley, C. J. & Johnson, K. W. 2013. Application of the PRECEDE-PROCEED Planning Model in Designing an Oral Health Strategy. *Journal of Theory and Practice in Dental Public Health*, 1.
- Birani, J., Santos, F. S., Scannavino, F. L. F. & Martins, A. T. 2008. Educational video on buccal health for care-takers of patients with special needs. *International Association of Disability and Oral Health*. Brazil: Journal of Disability and Oral Health.
- Bonardi, A., Clifford, C. J. & Fleming, C. K. 2018. Oral Health Care for Adults with IDD: A Summary of Evidence-based and Promising Practices Center for Developmental Disabilities Evaluation and Research. Massachusetts, USA: Eunice Kennedy Shriver Center.
- Bonetti, D., Hampson, V., Queen, K., Kirk, D., Clarkson, J. & Young, L. 2015. Improving oral hygiene for patients. *Nursing Standard*, 29, 44-50.
- Booth, A., Harris, J., Croot, E., Springett, J., Campbell, F. & Wilkins, E. 2013. Towards a methodology for cluster searching to provide conceptual and contextual "richness" for systematic reviews of complex interventions: case study (CLUSTER). *BMC Medical Research Methodology*, 13, 118.
- Borgnakke, W. S. 2015. Does Treatment of Periodontal Disease Influence Systemic Disease? *Dental Clinics of North America*, 59, 885-917.
- Brady, M., Furlanetto, D., Hunter, R. V., Lewis, S. & Milne, V. 2006. Staff-led interventions for improving oral hygiene in patients following stroke. *Cochrane Database Systematic Reviews*, Cd003864.
- Brehmer-Rinderer, B., Zigrovic, L. & Weber, G. 2014. Evaluating a health behaviour model for persons with and without an intellectual disability. *Journal of Intellectual Disability Research*, 58, 495-507.
- Broadbent, J. M., Thomson, W. M., Boyens, J. V. & Poulton, R. 2011. Dental plaque and oral health during the first 32 years of life. *Journal of the American Dental Association*, 142, 415-26.
- Brouwers, M., Kho, M.E., Browman, G.P, Cluzeau, F., Feder, G., Fervers, B., et al. 2011. AGREE II: Advancing guideline development, reporting and evaluation in healthcare. *Canadian Medical Association Journal* [Online].
- Brown, H. E., Atkin, A. J., Panter, J., Wong, G., Chinapaw, M. J. & Van Sluijs, E. M. 2016. Family-based interventions to increase physical activity in children: a systematic review, meta-analysis and realist synthesis. *Obesity Reviews*, 17, 345-60.
- Campanaro, M., Huebner, C. E. & Davis, B. E. 2014. Facilitators and barriers to twice daily tooth brushing among children with special health care needs. *Special Care in Dentistry*, 34, 185-192.
- Campbell, M. K., Piaggio, G., Elbourne, D. R. & Altman, D. G. 2012. Consort 2010 statement: extension to cluster randomised trials. *British Medical Journal: Research Methods and Reporting*, 345.
- Cane, J., Richardson, M., Johnston, M., Ladha, R. & Michie, S. 2015. From lists of behaviour change techniques (BCTs) to structured hierarchies: comparison of two methods of developing a hierarchy of BCTs. *British Journal of Health Psychology*, 20, 130-50.
- Care Alliance Ireland 2013. Family Caring in Ireland. Dublin: Care Alliance Ireland.
- Caton, S., Chadwick, D., Chapman, M., Turnbull, S., Mitchell, D. & Stansfield, J. 2012. Healthy lifestyles for adults with intellectual disability: Knowledge, barriers, and facilitators. *Journal of Intellectual & Developmental Disability*, 37, 248-259.
- Catteau, C., Mishellany, A., Faulks, D., Lassauzay, C. & Hennequin, M. 2011. Oral health status of persons with disabilities attending special establishments. First results of the national health education programme "Oral Health and

- Autonomy". *Bulletin du Groupement International pour la Recherche Scientifique en Stomatologie & Odontologie*, 50, 15-8.
- Central Statistics Office. 2006. *Census Volume 11: disability, carers and voluntary activities*. [Online]. Dublin: Stationery Office. Available: <https://www.cso.ie/en/census/census2006reports/census2006volume11-disabilitycarersandvoluntaryactivities/> [Accessed 21st December 2018].
- Central Statistics Office. 2016. *Census Profile 9 - Health, Disability and Carers* [Online]. Cork: Central Statistics Office. Available: <https://www.cso.ie/en/csolatestnews/presspages/2017/census2016profile9-healthdisabilityandcarers/> [Accessed 21st December 2018].
- Centre for Reviews and Dissemination 2008. *Systematic Reviews: CRD's guidance for undertaking reviews in health care*, University of York, UK, York Publishing Services Ltd.
- Chadwick, D. D., Mannan, H., Garcia Iriarte, E., McConkey, R., O'brien, P., Finlay, F., et al. 2013. Family voices: life for family carers of people with intellectual disabilities in Ireland. *Journal of Applied Research in Intellectual Disabilities*, 26, 119-32.
- Chadwick, D. D., Stubbs, J., Fovargue, S., Anderson, D., Stacey, G. & Tye, S. 2014. Training support staff to modify fluids to appropriate safe consistencies for adults with intellectual disabilities and dysphagia: an efficacy study. *Journal of Intellectual Disability Research*, 58, 84-98.
- Chestnutt, I. G., Schafer, F., Jacobson, A. P. M. & Stephen, K. W. 1998. The influence of toothbrushing frequency and post-brushing rinsing on caries experience in a caries clinical trial. *Community Dentistry and Oral Epidemiology*, 26, 406-411.
- Cochrane Effective Practice and Organisation of Care. 2017. Suggested risk of bias criteria for EPOC reviews. 2018. Available: <http://epoc.cochrane.org/resources/epoc-resources-review-authors>.
- Cochrane Oral Health Group 2018. Registered Titles, Protocols and Published Reviews. *Cochrane Oral Health*. <http://ohg.cochrane.org/oral-health-evidence>.
- Cooper, A. M., Coffey, M. & Dugdill, L. 2014. Challenges in designing, conducting, and reporting oral health behavioral intervention studies in primary school age children: methodological issues. *Pragmatic and Observational Research*, 5, 43-51.
- Cooper, A. M., O'Malley, L. A., Elison, S. N., Armstrong, R., Burnside, G., Adair, P., et al. 2013. Primary school-based behavioural interventions for preventing caries. *Cochrane Database of Systematic Reviews*.
- Covidence 2018. *Covidence systematic review software*. Melbourne, Australia: Veritas Health Innovation.
- Craig, P., Dieppe, P., MacIntyre, S., Michie, S., Nazareth, I., Petticrew, M., et al. 2008. Developing and evaluating complex interventions: the new Medical Research Council guidance. *British Medical Journal*, 337, a1655.
- Crowley, E., Whelton, H., Murphy, A., Kelleher, K., Cronin, M., Flannery, E., et al. 2005. Oral health of adults with an intellectual disability in residential care in Ireland 2003. Dublin: Dept. of Health and Children.
- Cumella, S., Ransford, N., Lyons, J. & Burnham, H. 2000. Needs for oral care among people with intellectual disability not in contact with Community Dental Services. *Journal of Intellectual Disability Research*, 44, 45-52.
- Dalkin, S. M., Greenhalgh, J., Jones, D., Cunningham, B. & Lhussier, M. 2015. What's in a mechanism? Development of a key concept in realist evaluation. *Implementation Science*, 10, 49.
- De Jongh, A., Van Houtem, C., Van Der Schoof, M., Resida, G. & Broers, D. 2008. Oral health status, treatment needs, and obstacles to dental care among noninstitutionalized children with severe mental disabilities in the Netherlands. *Special Care in Dentistry*, 28, 111-115.

- Deeks, J.J., Higgins, J.P.T. & Altman, D.G. 2011. Analysing data and undertaking meta-analyses. In: Higgins JPT & Green S. (eds.) *Cochrane Handbook for Systematic Reviews of Interventions*.: The Cochrane Collaboration
- Dougall, A. & Fiske, J. 2008a. Access to special care dentistry, part 1. Access. British Dental Journal, 204, 605-16.
- Dougall, A. & Fiske, J. 2008b. Access to special care dentistry, part 6. Special care dentistry services for young people. British Dental Journal, 205, 235-49.
- Dougall, A., Martinez Pereira, F., Molina, G., Eschevins, C., Daly, B. & Faulks, D. 2018. Identifying common factors of functioning, participation and environment amongst adults requiring specialist oral health care using the International Classification of Functioning, disability and health. *PloS one*, 13, e0199781-e0199781.
- Dwan, K., Altman, D. G., Clarke, M., Gamble, C., Higgins, J. P., Sterne, J. A., *et al.* 2014. Evidence for the selective reporting of analyses and discrepancies in clinical trials: a systematic review of cohort studies of clinical trials. *PLoS Medical*, 11, e1001666.
- Dye, B. A., Tan, S., Smith, V., Lewis, B. G., Barker, L. K., Thornton-Evans, G., *et al.* 2007. Trends in oral health status: United States, 1988-1994 and 1999-2004. *Vital Health Statistics 11*, 1-92.
- Egelberg, J. & Claffey, N. 1994. *Periodontal re-evaluation the scientific way*, Copenhagen, Munksgaard.
- Family Caregiver Alliance 2016. Caregiver Statistics: Demographics. *National Center on Caregiving*. USA.
- Faulks, D. & Hennequin, M. 2000. Evaluation of a long-term oral health program by carers of children and adults with intellectual disabilities. *Special Care in Dentistry*, 20, 199-208.
- Faulks, D., Norderyd, J., Molina, G., Mac Giolla Phadraig, C., Scagnet, G., Eschevins, C., *et al.* 2013. Using the International Classification of Functioning, Disability and Health (ICF) to Describe Children Referred to Special Care or Paediatric Dental Services. *PLOS ONE*, 8, e61993.
- Fernandez, J. B., Lim, L. J., Dougherty, N., Lasasso, J., Atar, M. & Daronch, M. 2012. Oral health findings in athletes with intellectual disabilities at the NYC Special Olympics. *Special Care in Dentistry*, 32, 205-9.
- Fleiss, J.L, Park, M.H, Chilton, N.W, Alman, J.E, Feldman, R.S & Chauncey, H.H. 1987. Representativeness of the "Ramfjord teeth" for epidemiologic studies of gingivitis and periodontitis. *Community Dentistry and Oral Epidemiology* 15, 221 - 224.
- Fletcher, A., Jamal, F., Moore, G., Evans, R. E., Murphy, S. & Bonell, C. 2016. Realist complex intervention science: Applying realist principles across all phases of the Medical Research Council framework for developing and evaluating complex interventions. *Evaluation (London, England: 1995)*, 22, 286-303.
- Forsell, M. S., P.; Kullberg, E.; Johansson, O.; Wedel, P.; Herbst, B.; Hoogstraate, J. 2011. Attitudes and perceptions towards oral hygiene tasks among geriatric nursing home staff. *International Journal of Dental Hygiene*, 9, 199-203.
- Fujita, M., Ogawa, M., Nagasawa, A. & Tomioka, I. 2015. Development of International Classification of Functioning, Disability and Health (ICF) Core Sets for Use With People With Severe Intellectual Disabilities. *The Japanese Journal of Special Education*, 53, 97-105.
- Gabre, P., Martinsson, T. & Gahnberg, L. 2001. Longitudinal study of dental caries, tooth mortality and interproximal bone loss in adults with intellectual disability. *European Journal of Oral Sciences*, 109, 20-6.
- Garmyn, P., Van Steenberghe, D. & Quirynen, M. 1998. Efficacy of plaque control in the maintenance of gingival health: Plaque control in primary and secondary prevention. In: Lang, N. P., Attström, R. & Löe, H. (eds.) *Proceedings of the European Workshop on Mechanical Plaque Control*. Berlin: Quintessence Pub. Inc.

- Glassman, P. & Miller, C. 2009. Social supports and prevention strategies as adjuncts and alternatives to sedation and anesthesia for people with special needs. *Special Care in Dentistry*, 29, 31-8.
- Glassman, P. & Miller, C. E. 2003. Preventing dental disease for people with special needs: the need for practical preventive protocols for use in community settings. *Special Care in Dentistry*, 23, 165-167.
- Glassman, P. & Miller, C. E. 2006. Effect of preventive dentistry training program for caregivers in community facilities on caregiver and client behavior and client oral hygiene. *New York State Dental Journal*, 72, 38-46.
- Glick, M., Williams, D. M., Kleinman, D. V., Vujicic, M., Watt, R. G. & Weyant, R. J. 2016. A new definition for oral health developed by the FDI World Dental Federation opens the door to a universal definition of oral health. *International Dental Journal*, 66, 322-324.
- Gordon, S. M., Dionne, R. A. & Snyder, J. 1998. Dental fear and anxiety as a barrier to accessing oral health care among patients with special health care needs. *Special Care in Dentistry*, 18, 88-92.
- Graham, I. D., Logan, J., Harrison, M. B., Straus, S. E., Tetroe, J. & Caswell, W. 2006. Lost in knowledge translation: time for a map? *Journal of Continuing Education in Health Professions*, 26.
- Grant, M. & Booth, A. 2009. A typology of reviews: an analysis of 14 review types and associated methodologies. *Health Information and Libraries Journal*, 26.
- Greene, J. G. & Vermillion, J. R. 1960. The oral hygiene index: a method for classifying oral hygiene status. *The Journal of the American Dental Association*, 61, 172-179.
- Greenhalgh, T., Kristjansson, E. & Robinson, V. 2007. Realist review to understand the efficacy of school feeding programmes. *British Medical Journal*, 335.
- Health Research Board 2017. Annual Report of the National Intellectual Disability Database Committee 2016 *HRB Statistics Series*. Dublin: Health Research Board.
- Hennequin, M., Moysan, V., Jourdan, D., Dorin, M. & Nicolas, E. 2008. Inequalities in oral health for children with disabilities: a French national survey in special schools. *PLoS One*, 3, e2564.
- Higgins, J. & Green, S. 2011. Cochrane Handbook for Systematic Reviews of Interventions Version 5.1.0 [updated March 2011]. *The Cochrane Collaboration*. Available from <http://handbook.cochrane.org>.
- Higgins, J.P.T., Altman, D.G. & Sterne, J.A.C. 2011a. Assessing risk of bias in included studies. In: Higgins JPT & Green S (eds.) *Cochrane Handbook for Systematic Reviews of Interventions*. The Cochrane Collaboration.
- Higgins, J.P.T., Deeks, J.J. & Altman, D.G. 2011b. Special topics in statistics. In: Higgins JPT & Green S (eds.) *Cochrane Handbook for Systematic Reviews of Interventions*.: The Cochrane Collaboration.
- Hithersay, R., Strydom, A., Moulster, G. & Buszewicz, M. 2014. Carer-led health interventions to monitor, promote and improve the health of adults with intellectual disabilities in the community: A systematic review. *Research in Developmental Disabilities*, 35, 887-907.
- Hopewell, S., Clarke, M. J., Stewart, L. & Tierney, J. 2007. Time to publication for results of clinical trials. *The Cochrane Library*.
- Hopewell, S., Loudon, K., Clarke, M. J., Oxman, A. D. & Dickersin, K. 2009. Publication bias in clinical trials due to statistical significance or direction of trial results. *The Cochrane Library*.
- Horsley, T., Dingwall, O. & Sampson, M. 2011. Checking reference lists to find additional studies for systematic reviews. *The Cochrane Library*.
- Horwitz, S. M., Kerker, B. D., Owens, P. L. & Zigler, E. 2000. The Health Status and Needs of Individuals with Mental Retardation. New Haven CT: Yale University School of Medicine and Special Olympics, Inc.

- Hujoel, P. P., Cunha-Cruz, J., Loesche, W. J. & Robertson, P. B. 2005. Personal oral hygiene and chronic periodontitis: a systematic review. *Periodontology* 2000, 37, 29-34.
- Johnson, I. G. 2013. Nurses' emotions and oral care for hospitalised adults. Doctor of Philosophy PhD, Cardiff University.
- Kalf-Scholte, S., Van Der Weijden, G., Bakker, E. & Slot, D. 2017. Plaque removal with triple-headed vs single-headed manual toothbrushes—a systematic review—. *International Journal of Dental Hygiene*.
- Kastner, M., Tricco, A. C., Soobiah, C., Lillie, E., Perrier, L., Horsley, T., *et al.* 2012. What is the most appropriate knowledge synthesis method to conduct a review? Protocol for a scoping review. *BMC Medical Research Methodology*, 12, 114.
- Kay, E. & Locker, D. 1998. A systematic review of the effectiveness of health promotion aimed at improving oral health. *Community Dental Health*, 15, 132-44.
- Kaye, P. L., Fiske, J., Bower, E. J., Newton, J. T. & Fenlon, M. 2005. Views and experiences of parents and siblings of adults with Down Syndrome regarding oral healthcare: a qualitative and quantitative study. *British Dental Journal*, 198, 571-578.
- Kobayashi, L. Y. & Ash, M. 1964. A clinical evaluation of an electric toothbrush used by orthodontic patients. *Clinical Evaluation*, 34, 209 - 219.
- Kuijken, N. M. J., Naaldenberg, J., Nijhuis-Van Der Sanden, M. W. & De Valk, H. 2016. Healthy living according to adults with intellectual disabilities: towards tailoring health promotion initiatives. *Journal of Intellectual Disability Research*, 60, 228-241.
- Kurbasic, I., Pandza, H., Masic, I., Huseinagic, S., Tandir, S., Alicajic, F., *et al.* 2008. The advantages and limitations of international classification of diseases, injuries and causes of death from aspect of existing health care system of Bosnia and Herzegovina. *Acta Informatica Medica*, 16, 159-61.
- Lader, D., Chadwick, B., Chestnutt, I., Harker, R., Morris, J., Nuttall, N., *et al.* 2005. Children's dental health in the United Kingdom, 2003. London: Office for National Statistics.
- Lamont, T. J., Clarkson, J. E., Ricketts, D. N. J., Heasman, P. A. & Ramsay, C. R. 2017. Core outcomes in periodontal trials: study protocol for core outcome set development. *Trials*, 18, 436.
- Lang, N. P., Schätzle, M. A. & Löe, H. 2009. Gingivitis as a risk factor in periodontal disease. *Journal of Clinical Periodontology*, 36, 3-8.
- Levac, D., Colquhoun, H. & O'Brien, K. K. 2010. Scoping studies: advancing the methodology. *Implementation Science*, 5, 1-9.
- Lobene, R. R., Weatherford, T., Ross, N. M., Lamm, R. A. & Menaker, L. 1986. A modified gingival index for use in clinical trials. *Clinical Preventive Dentistry*, 8, 3-6.
- Löe, H. 1967. The Gingival Index, the Plaque Index and the Retention Index Systems. *Journal of Periodontology*, 38, 610-616.
- Löe, H. 2000. Oral hygiene in the prevention of caries and periodontal disease. *International Dental Journal*, 50, 129-139.
- Löe, H., Theilade, E. & Jensen, S. 1965. Experimental gingivitis in man. *Journal of Periodontology*, 36, 177-87.
- Loudon, K., Zwarenstein, M., Sullivan, F., Donnan, P. & Treweek, S. 2013. Making clinical trials more relevant: improving and validating the PRECIS tool for matching trial design decisions to trial purpose. *Trials*, 14, 115.
- Mac Giolla Phadraig, C., Guerin, S. & Nunn, J. 2013. Train the trainer? A randomized controlled trial of a multi-tiered oral health education programme in community-based residential services for adults with intellectual disability. *Community Dental and Oral Epidemiology* 41, 182-92.
- Mac Giolla Phadraig, C., McCallion, P., Cleary, E., McGlinchey, E., Burke, E., McCarron, M., *et al.* 2015. Total tooth loss and complete denture use in older

- adults with intellectual disabilities in Ireland. *Journal of Public Health Dentistry*, 75, 101-108.
- Mac Giolla Phadraig, C., Nunn, J., McCallion, P. & McCarron, M. 2018. Prevalence of edentulism among adults with intellectual disabilities: A narrative review informed by systematic review principles. *Special Care in Dentistry*, 38, 191-200.
- Marcenes, W., Wright, D., Murray, S., Redshaw, P. & Bennett, U. 2008. The oral health of three-four-year-old children in Inner North East London in 2007. London: The Institute of Dentistry, Barts and the London School of Medicine and Dentistry, Queen Mary University of London, Hackney PCT, Tower Hamlets PCT, Newham PCT.
- Marks, L., Wong, A., Perlman, S., Shellard, A. & Fernandez, C. 2017. Global oral health status of athletes with intellectual disabilities. *Clinical Oral Investigations*.
- McAlister, T. & Bradley, C. 2003. The oral and dental health of children in special national schools in the Eastern Region Health Authority, Ireland 1999/2000. *Journal of Disability and Oral Health*, 4, 69-72.
- McMahon, T. & Ward, P. R. 2012. HIV among immigrants living in high-income countries: a realist review of evidence to guide targeted approaches to behavioural HIV prevention. *Systematic Reviews*, 1, 56.
- Michie, S. & Abraham, C. 2004. Interventions to change health behaviours: evidence-based or evidence-inspired? *Psychology & Health*, 19, 29-49.
- Michie, S., Johnston, M., Francis, J., Hardeman, W. & Eccles, M. 2008. From theory to intervention: mapping theoretically derived behavioural determinants to behaviour change techniques. *Applied Psychology*, 57, 660 - 680.
- Michie, S., Richardson, M., Johnston, M., Abraham, C., Francis, J., Hardeman, W., et al. 2013. The behavior change technique taxonomy (v1) of 93 hierarchically clustered techniques: building an international consensus for the reporting of behavior change interventions. *Annals of Behavioural Medicine*, 46, 81-95.
- Michie, S., Van Stralen, M. M. & West, R. 2011. The behaviour change wheel: a new method for characterising and designing behaviour change interventions. *Implementation Science*, 6, 42.
- Minihan, P. M., Morgan, J. P., Park, A., Yantsides, K. E., Nobles, C. J., Finkelman, M. D., et al. 2014. At-home oral care for adults with developmental disabilities: a survey of caregivers. *Journal of the American Dental Association*, 145, 1018-25.
- Moher, D., Schulz, K. F. & Altman, D. G. 2001. The CONSORT statement: revised recommendations for improving the quality of reports of parallel-group randomised trials. *The Lancet*, 357, 1191-1194.
- Mombelli, A. W. 1998. The role of dental plaque in the initiation and progression of periodontal diseases. In: Lang, N. P., Attström, R. & Löe, H. (eds.) *Proceeding of the European Workshop on Mechanical Plaque Control*. Berlin: Quintessence Publishing Co. Inc.
- Moreira, M. J. S., Schwertner, C., Jardim, J. J. & Hashizume, L. N. 2016. Dental caries in individuals with Down syndrome: a systematic review. *International Journal of Paediatric Dentistry*, 26, 3-12.
- Morgan, J. P., Minihan, P. M., Stark, P. C., Finkelman, M. D., Yantsides, K. E., Park, A., et al. 2012. The oral health status of 4,732 adults with intellectual and developmental disabilities. *Journal of the American Dental Association*, 143, 838-46.
- National Disability Survey 2008. *National Disability Survey 2006: first results* Dublin, Stationery Office.
- Newton, J. T. & Asimakopoulou, K. 2015. Managing oral hygiene as a risk factor for periodontal disease: a systematic review of psychological approaches to behaviour change for improved plaque control in periodontal management. *Journal of Clinical Periodontology*, 42, S36-S46.

- Niemeier, B. S., Hektner, J. M. & Enger, K. B. 2012. Parent participation in weight-related health interventions for children and adolescents: A systematic review and meta-analysis. *Preventive Medicine*, 55, 3-13.
- Novak, I., Hines, M., Goldsmith, S. & Barclay, R. 2012. Clinical prognostic messages from a systematic review on cerebral palsy. *Pediatrics*, 130, e1285-312.
- Nutbeam, D. 1998. Evaluating Health Promotion—Progress, Problems and solutions. *Health Promotion International*, 13, 27-44.
- NVivo 11 2012. *NVivo qualitative data analysis Software*, QSR International Pty Ltd.
- Office of Disease Prevention and Health Promotion. 2018. *Healthy People 2020* [Online]. Washington DC Office of Disease Prevention and Health Promotion [Accessed 29th November 2018].
- Oliveira, J. S., Prado Júnior, R. R., De Sousa Lima, K. R., De Oliveira Amaral, H., Moita Neto, J. M. & Mendes, R. F. 2013. Intellectual disability and impact on oral health: a paired study. *Special Care in Dentistry*, 33, 262-268.
- O'Mullane, D. 1976. Efficiency in clinical trials of caries preventive agents and methods. *Community Dental and Oral Epidemiology*, 4, 190-4.
- O'Mullane, D., James, P., Whelton, H. & Parnell, C. 2012. Methodological issues in oral health research: intervention studies. *Community Dentistry and Oral Epidemiology*, 40, 15-20.
- Owens, J. 2011. Oral health promotion for children with disabilities in the Republic of Ireland: independent qualitative evaluation of a multi-sectoral oral health promotion intervention for children with disabilities. *Global Health Promotion*, 18, 69-71.
- Parnell, C., Connolly, E., O'Farrell, M., Cronin, M., Flannery, E. & Whelton, H. 2007. Oral Health of 5-year-old children in the North East, 2002. In: Oral Health Services Research Centre and HSE Dublin North East (ed.). Dublin.
- Pawson, R. 2006. *Evidence-based Policy: A realist perspective*, London, SAGE Publications Ltd.
- Pawson, R. 2013. *The Science of Evaluation: A Realist Manifesto*, London, Sage Publications Ltd.
- Pawson, R. Evidenced-Based Medicine and Evidenced-Based Policy: The World's Most Perfectly Developed Method and the 79 Pound Weakling. 2nd International Conference on Realist Evaluation and Synthesis, 2016 Barbican Centre London, UK.
- Pawson, R., Greenhalgh, J., Brennan, C. & Glidewell, E. 2014. Do reviews of healthcare interventions teach us how to improve healthcare systems? *Social Science & Medicine*, 114, 129-137.
- Pawson, R., Greenhalgh, T., Harvey, G. & Walshe, K. 2005. Realist review—a new method of systematic review designed for complex policy interventions. *Journal of Health Services Research and Policy*, 10.
- Pawson, R. & Tilley, N. 1996. What's Crucial in Evaluation Research: A Reply to Bennett. *British Journal of Criminology*, 36, 574-578.
- Pawson, R. & Tilley, N. 2004. Realist Evaluation. UK: British Cabinet Office.
- Pearson, M., Chilton, R., Woods, H. B., Wyatt, K., Ford, T., Abraham, C., et al. 2012. Implementing health promotion in schools: protocol for a realist systematic review of research and experience in the United Kingdom (UK). *Systematic reviews*, 1, 48.
- Pearson, M., Hunt, H., Cooper, C., Shepperd, S., Pawson, R. & Anderson, R. 2015. Providing effective and preferred care closer to home: a realist review of intermediate care. *Health and Social Care in the Community*, 23, 577-93.
- Petersen, P. E. 2003. The World Oral Health Report 2003: continuous improvement of oral health in the 21st century – the approach of the WHO Global Oral Health Programme. *Community Dentistry and Oral Epidemiology*, 31, 3-24.
- Petersen, P. E. & Baez, R. J. 2013. *Oral health surveys: basic methods*. 1. Oral health surveys. 2. Dental health. 3. Data collection. 4. Oral health surveillance.

- Petrovic B. B., Peric T.O., Markovic D. L. J., Bajkin B. B., Petrovic, D., Blagojevic, B., *et al.* 2016. Unmet oral health needs among persons with intellectual disability. *Research in developmental disabilities*, 59, 370-377.
- Petticrew, M., Anderson, L., Elder, R., Grimshaw, J., Hopkins, D., Hahn, R., *et al.* 2015. Complex interventions and their implications for systematic reviews: A pragmatic approach. *International Journal of Nursing Studies*, 52, 1211-1216.
- Prabhu, N. T., Nunn, J. H., Evans, D. J. & Girdler, N. M. 2010. Access to dental care - Parents' and caregivers' views on dental treatment services for people with disabilities. *Special Care in Dentistry*, 30, 35-45.
- Pradhan, A., Slade, G. D. & Spencer, A. J. 2009. Access to dental care among adults with physical and intellectual disabilities: residence factors. *Australian Dental Journal*, 54, 204-11.
- Prangnell, S. J. & Green, K. 2008. A cognitive behavioural intervention for dental anxiety for people with learning disabilities: a case study. *British Journal of Learning Disabilities*, 36, 242-248.
- Public Health England. 2015. Child Dental Health Survey 2013, England, Wales and Northern Ireland [Online]. Available: <https://digital.nhs.uk/data-and-information/publications/statistical/children-s-dental-health-survey/child-dental-health-survey-2013-england-wales-and-northern-ireland> [Accessed 27 January 2019].
- Public Health England 2015. Oral health survey of five-year-old and 12-year-old children attending special support schools 2014. *Public dental health epidemiology programme*. London.
- Public Health England 2017. Dental health among adults with learning disabilities in England. In: Department of Health (ed.) *Fact Sheet*. London, UK.
- Rawlinson, S. R. 2001. The Dental and Oral Care Needs of Adults with a Learning Disability Living in a Rural Community: Consideration of the Issues. *Journal of Intellectual Disabilities*, 5, 133-156.
- Redding, C. A., Rossi, J. S., Rossi, S. R., Velicer, W. F. & Prochaska, J. O. Health behavior models. *International Electronic Journal of Health Education*, 2000. Citeseer.
- Renz, A., Ide, M., Newton, T., Robinson, P. & Smith, D. 2007. Psychological interventions to improve adherence to oral hygiene instructions in adults with periodontal diseases. *Cochrane Database of Systematic Reviews*.
- Round, R., Marshall, B. & Horton, K. 2005. Planning for effective health promotion evaluation. In: Department of Human Services (ed.). Melbourne: Victorian Government
- Rycroft-Malone, J., McCormack, B., Hutchinson, A. M., Decorby, K., Bucknall, T. K., Kent, B., *et al.* 2012. Realist synthesis: illustrating the method for implementation research. *Implementation Science*, 7, 33.
- Sagheri, D., Mcloughlin, J. & Nunn, J. H. 2013. Dental caries experience and barriers to care in young children with disabilities in Ireland. *Quintessence International*, 44, 159-69.
- Saxer Up & Mühlemann Hr 1975. Motivation and education. *SSO Schweiz Monatsschr Zahnheilkd*, 85, 9005-19.
- Schalock, R. L., Borthwick-Duffy, S. A., Bradley, V. J., Buntinx, W. H. E., Coulter, D. L., Craig, E. M., *et al.* 2010. Intellectual Disability: Definition, Classification, and Systems of Supports. Eleventh Edition. American Association on Intellectual and Developmental Disabilities.
- Schou, L. 1998. Behavioral aspects of dental plaque control measures: An oral health promotion perspective. In: Lang, N. P. & Attström, R., Harald (eds.) *Proceedings of the European Workshop on Mechanical Plaque Control*. Berlin: Quintessence Pub. Inc.
- Schulz, K. F., Altman, D. G. & Moher, D. 2010. CONSORT 2010 Statement: updated guidelines for reporting parallel group randomised trials. *British Medical Journal*, 340.

- Schünemann, H., Brożek, J., Guyatt, G. & Oxman, A. 2013. *Handbook for grading the quality of evidence and the strength of recommendations using the GRADE approach*. [Online]. Available: <http://gdt.guidelinedevelopment.org/app/handbook/handbook.html#h.52ccy41iwbbon> [Accessed 21st December 2018].
- Schwarzer, R. 2008. Modelling health behavior change: How to predict and modify the adoption and maintenance of health behaviors. *Applied Psychology*, 57, 1-29.
- Schwarzer, R., Lippke, S. & Luszczynska, A. 2011. Mechanisms of health behavior change in persons with chronic illness or disability: the Health Action Process Approach (HAPA). *Rehabilitation Psychology*, 56, 161.
- Schwendicke, F., Lamont, T. & Innes, N. 2015. Outcomes in Trials for Management of Caries Lesions (OuTMaC): protocol. *Trials*, 16, 397.
- Scottish Government 2010. Caring together; Carers Strategy for Scotland 2010 - 2015. In: Government, S. (ed.) *Healthier Scotland*. Edinburgh.
- Shaw, L. & Murray, J. J. 1977. A new index for measuring extrinsic stain in clinical trials. *Community Dental and Oral Epidemiology*, 5, 116-120.
- Skinner, B. F. 1969. *Contingencies of reinforcement: A theoretical analysis*, USA, Meredith Corporation.
- Sniehotta, F. F. 2009. Towards a theory of intentional behaviour change: Plans, planning, and self-regulation. *British Journal of Health Psychology*, 14, 261-273.
- Sniehotta, F. F., Nagy, G., Scholz, U. & Schwarzer, R. 2006. The role of action control in implementing intentions during the first weeks of behaviour change. *British Journal of Social Psychology*, 45, 87-106.
- Stapleton, S. 2015. *How can we impact on the oral health status of 0-6-year-old children with disabilities in Ireland?* Degree of Professional Doctorate in Special Care Dentistry, Trinity College Dublin.
- Sterne, J. A., Hernán, M. A., Reeves, B. C., Savović, J., Berkman, N. D., Viswanathan, M., et al. 2016. ROBINS-I: a tool for assessing risk of bias in non-randomised studies of interventions. *British Medical Journal*, 355.
- Stewart, J., Wolfe, G., Maeder, L. & Hartz, G. 1996. Changes in dental knowledge and self-efficacy scores following interventions to change oral hygiene behavior. *Patient Education and Counselling*, 26, 269-277.
- Stewart, K., Gill, P., Chadwick, B. & Treasure, E. 2008. Qualitative research in dentistry. *British Dental Journal*, 204, 235.
- Taylor-Dillon, F., Friel, S., Sixsmith, J. & Kelleher, C. 2003. A Review of Oral Health Promotion / Education Activity in the Republic of Ireland and A Study of Attitudes, Knowledge and Behaviour of Special Needs Groups Towards Oral Health. In: Centre for Health Promotion Studies (ed.). Galway.
- Tedesco, L. A., Keffer, M. A. & Fleck-Kandath, C. 1991. Self-efficacy, reasoned action, and oral health behavior reports: A social cognitive approach to compliance. *Journal of Behavioral Medicine*, 14, 341-355.
- Thole, K., Chalmers, J., Ettinger, R. L. & Warren, J. 2010. Iowa intermediate care facilities: An evaluation of care providers' attitudes toward oral hygiene care. *Special Care in Dentistry*, 30, 99-105.
- Thorpe, K. E., Zwarenstein, M., Oxman, A. D., Treweek, S., Furberg, C. D., Altman, D. G., et al. 2009. A pragmatic-explanatory continuum indicator summary (PRECIS): a tool to help trial designers. *Journal of Clinical Epidemiology*, 62, 464-75.
- Tiller, S., Wilson, K. I. & Gallagher, J. E. 2001. Oral health status and dental service use of adults with learning disabilities living in residential institutions and in the community. *Community Dental Health*, 18, 167-171.
- Tricco, A. C., Antony, J., Soobiah, C., Kastner, M., Cogo, E., MacDonald, H., et al. 2016a. Knowledge synthesis methods for generating or refining theory: a scoping review reveals that little guidance is available. *Journal of Clinical Epidemiology*, 73, 36-42.

- Tricco, A. C., Soobiah, C., Antony, J., Cogo, E., MacDonald, H., Lillie, E., *et al.* 2016b. A scoping review identifies multiple emerging knowledge synthesis methods, but few studies operationalize the method. *Journal of Clinical Epidemiology*, 73, 19-28.
- Turesky, S., Gilmore, N. D. & Glickman, I. 1970. Reduced Plaque Formation by the Chloromethyl Analogue of Vitamin C. *Journal of Periodontology*, 41, 41-43.
- Turner, S., Sweeney, M., Kennedy, C. & MacPherson, L. 2008. The oral health of people with intellectual disability participating in the UK Special Olympics. *Journal of Intellectual Disability Research*, 52, 29-36.
- Vale, M. D. C., Pereira-Da-Silva, L., Pimentel, M. J., Marques, T. N., Rodrigues, H., Cunha, G., *et al.* 2017. Classifying Functioning of Children and Adolescents with Intellectual Disability: The Utility of the International Classification of Functioning, Disability and Health for Children and Youth. *Journal of Policy & Practice in Intellectual Disabilities*, 14, 285-292.
- Van Der Weijden, F. & Slot, D. E. 2011. Oral hygiene in the prevention of periodontal diseases: the evidence. *Periodontology 2000*, 55, 104-123.
- Waldron, C., Mac Giolla Phadraig, C., Comiskey, C. & Nunn, J. 2016. Results of a Scoping Review of Oral Health Interventions for Children or Adolescents with Disabilities. *Journal of Disability & Oral Health*, 17, 144 - 149.
- Wallace, J., Byrne, C. & Clarke, M. 2014. Improving the uptake of systematic reviews: a systematic review of intervention effectiveness and relevance. *BMJ Open*, 4, e005834-e005834.
- Watt, R. G., Harnett, R., Daly, B., Fuller, S. S., Kay, E., Morgan, A., *et al.* 2006. Evaluating oral health promotion: need for quality outcome measures. *Community Dental and Oral Epidemiology*, 34, 11-7.
- Watt, R. G. & Marinho, V. C. 2005. Does oral health promotion improve oral hygiene and gingival health? *Periodontology 2000*, 37, 35-47.
- Webb, T. L. & Sheeran, P. 2008. Mechanisms of implementation intention effects: The role of goal intentions, self-efficacy, and accessibility of plan components. *British Journal of Social Psychology*, 47, 373-395.
- Wei, S. & Lang, K. P. 1981. Periodontal epidemiological indices for children and adolescents: I. Gingival and periodontal health assessments. *Pediatric Dentistry*, 3, 353-60.
- Wei, S. H. Y. & Lang, N. P. 1982. Periodontal epidemiological indices for children and adolescents: II. Evaluation of oral hygiene; III. Clinical applications. *The American Academy of Pedodontics*, 4, 64-73.
- Whelton, H., Crowley, E., Nunn, J., Murphy, A., Kelleher, V., Guiney, H., *et al.* 2009. Oral health of children attending special needs schools and day care centres. Cork University Hospital.
- Whelton, H., Crowley, E., O'Mullane, D., Harding, M., Guiney, H., Cronin, M., *et al.* 2006. North South Survey of Children's Oral Health in Ireland 2002. Department of Health and Children, Dublin.
- Whelton, H., Crowley, E., O'Mullane, D., Woods, N., McGrath, C., Kelleher, V., *et al.* 2007. Oral Health of Irish Adults 2000 – 2002. Dublin: Department of Health and Children, Dublin.
- Willems, M., Hilgenkamp, T. I. M., Havik, E., Waninge, A. & Melville, C. A. 2017. Use of behaviour change techniques in lifestyle change interventions for people with intellectual disabilities: A systematic review. *Research in Developmental Disabilities*, 60, 256-268.
- Williamson, P. R., Altman, D. G., Bagley, H., Barnes, K. L., Blazeby, J. M., Brookes, S. T., *et al.* 2017. The COMET Handbook: version 1.0. *Trials*, 18, 280-280.
- Wong, G. 2018. Data gathering in realist reviews: searching for needles in haystacks. In: Emmel, N., Greenhalgh, J., Manzano, A., Monaghan, M. & Dalkin, S. (eds.) *Doing Realist Research*. London: Sage.
- Wong, G., Greenhalgh, T., Westhorp, G., Buckingham, J. & Pawson, R. 2013. RAMESES publication standards: realist syntheses. *BMC Medical* 11, 21.

- World Health Organization 1978. Epidemiology, etiology and prevention of periodontal diseases. *World Health Organisation*. Geneva: Scientific Group on Epidemiology etiology and prevention of periodontal diseases.
- World Health Organisation. 1992. *International statistical Classification of Diseases and related health problems tenth revision (ICD-10)* [Online]. Geneva, Switzerland: WHO. Available: <https://icd.who.int/browse10/2016/en#/F70-F79> [Accessed 31st December 2018].
- World Health Organization 2001. The International Classification of Functioning, Disability and Health. *World Health Organisation*. Geneva, Switzerland.
- World Health Organization 2011. World report on disability. Geneva, Switzerland: WHO Press.
- Worthington, H., Clarkson, J. & Weldon, J. 2015. Priority oral health research identification for clinical decision-making. *Evidence-based dentistry*, 16, 69-71.
- Worthington, H. V., Clarkson, J. E., Bryan, G. & Beirne, P. V. 2013. Routine scale and polish for periodontal health in adults. *Cochrane Database of Systematic Reviews*.
- Yaacob, M., Worthington, H. V., Deacon, S. A., Deery, C., Walmsley, A. D., Robinson, P. G., et al. 2014. Powered versus manual toothbrushing for oral health. *Cochrane Database of Systematic Reviews*. John Wiley & Sons, Ltd.
- Young, T. & Hopewell, S. 2011. Methods for obtaining unpublished data. *Cochrane Database of Systematic Reviews*
- Zaihan, O., Normastura, A., Azizah, Y. & Khairi, M. 2015. Plaque Maturity and Problems Encountered by Mothers During Tooth Brushing Among Down Syndrome Children in The Northeast of Peninsular Malaysia. *International Journal of Public Health and Clinical Sciences*, 2, 159-173.
- Zeller, A. 2004. *Klinische Untersuchung der Putzeffektivität und Handhabbarkeit einer elektronischen Zahnbürste, einer dreiköpfigen Handzahnbürste und einer konventionellen Handzahnbürste bei Patienten mit Behinderungen / Clinical investigation of the brushing effectiveness and manageability of an electronic toothbrush, a three-headed manual toothbrush and a conventional manual toothbrush in patients with disabilities.*, Freie Universität Berlin
- Zhou, N., Wong, H. M., Wen, Y. F. & McGrath, C. 2017. Oral health status of children and adolescents with intellectual disabilities: a systematic review and meta-analysis. *Developmental Medicine and Child Neurology*, 59, 1019-1026.
- Zhou, N., Wong, H. M., Wen, Y. F. & McGrath, C. 2018. Efficacy of caries and gingivitis prevention strategies among children and adolescents with intellectual disabilities: a systematic review and meta-analysis. *Journal of Intellectual Disability Research*. (Early view: Online Version of Record before inclusion in an issue).
- Zimmermann, H., Zimmermann, N., Hagenfeld, D., Veile, A., Kim, T.-S. & Becher, H. 2015. Is frequency of tooth brushing a risk factor for periodontitis? A systematic review and meta-analysis. *Community Dentistry and Oral Epidemiology*, 43, 116-127.

7.2 Scoping Review: Included Studies

- Adelson, J. J. 1965. The effects of dental treatment on behavior of handicapped patients. *Journal of the American Dental Association*, 71, 1411-5.
- Adiwoso, A. S. & Pilot, T. 1999. Results of oral health and hygiene education in an institution for multiple handicapped children in Indonesia. *International Dental Journal*, 49, 82-9.
- Ahmadi, M., Abbasi, M. & Bahaadinbeigy, K. 2015. Design and implementation of a software for teaching health related topics to deaf students: The first experience in Iran. *Acta Informatica Medica*, 23, 76-80.

- Alacam, A. & Kolcuoğlu, N. 2007. Effects of two types of appliances on orofacial dysfunctions of disabled children. *British Journal of Developmental Disabilities*, 53, 111-123.
- Albino, J. E., Schwartz, B. H., Goldberg, H. J. & Stern, M. E. 1979. Results of an oral hygiene program for severely retarded children. *ASDC Journal of Dentistry for Children*, 46, 25-28.
- Attrill, M. & Hobson, P. 1984. The organization of dental care for groups of medically handicapped children. *Community dental health*, 1, 21-27.
- Aved, B. M., Meyers, L. S. & Burmas, E. L. 2008. Increasing dental care for very young children: what can training accomplish? *Journal of the California Dental Association*, 36, 931-940.
- Baens-Ferrer, C., Roseman, M. M., Dumas, H. M. & Haley, S. M. 2005. Parental perceptions of oral health-related quality of life for children with special needs: impact of oral rehabilitation under general anesthesia. *Pediatric Dentistry*, 27, 137-142.
- Balzer, J. 2007. Improving systems of care for people with special needs: the ASTDD Best Practices Project. *Pediatric dentistry*, 29, 123-128.
- Barber, S. 2011. Smile Mumias: oral health education and dental care for children in Mumias District, Western Kenya (formerly the Akhonya Dental Project): one year on. *British Dental Journal*, 211, 43-46.
- Barker, G. J., Call, S. K. & Gamis, A. S. 1995. Oral care with vancomycin paste for reduction in incidence of alpha-hemolytic streptococcal sepsis. *Journal of Pediatric Hematology & Oncology*, 17, 151-5.
- Barnett, M. L., Ziring, P., Friedman, D. & Sonnenberg, E. M. 1988. Dental treatment program for patients with mental retardation. *Mental Retardation*, 26, 310-3.
- Bay, L. M. & Russel, B. G. 1975. Effect of chlorhexidine on dental plaque and gingivitis in mentally retarded children. *Community Dentistry and Oral Epidemiology*, 3, 267-270.
- Baygin, O., Tuzuner, T., Kusgoz, A., Senel, A. C., Tanriver, M. & Arslan, I. 2014. Antibacterial effects of fluoride varnish compared with chlorhexidine plus fluoride in disabled children. *Oral Health & Preventive Dentistry*, 12, 373-82.
- Bishop, M. R., Kenzer, A. L., Coffman, C. M., Tarbox, C. M., Tarbox, J. & Lanagan, T. M. 2013. Using stimulus fading without escape extinction to increase compliance with toothbrushing in children with autism. *Research in Autism Spectrum Disorders*, 7, 680-686.
- Bizarra, F. & Ribeiro, S. 2009. Improving toothbrushing behaviour in an institution for the disabled in Lisbon, Portugal. *International Journal of Dental Hygiene*, 7, 182-187.
- Blaine, E., Bell, J. & Smith, J. 1971. Oral hygiene supplement for handicapped children. *Dental Survey*, 47, 29-31.
- Borutta, A. & Heinrich, R. 1992. Long-term results with dental treatment concepts for disabled children]. [German. *Deutsche Zahnärztliche Zeitschrift*, 47, 23-25.
- Bothwell, J. E. C., K.; Dooley, J. M.; Gordon, K. E.; Anderson, R.; Wood, E. P.; Camfield, C. S.; Camfield, P. R. 2002. Botulinum toxin A as a treatment for excessive drooling in children. *Pediatric Neurology*, 27, 18-22.
- Brayer, L., Goultschin, J. & Mor, C. 1985. The effect of chlorhexidine mouthrinses on dental plaque and gingivitis in mentally retarded individuals. *Clinical Preventive Dentistry*, 7, 26-28.
- Brown, J. P. 1975. Dental treatment for handicapped patients. I. The efficacy of a preventive programme for children. II. Economics of dental treatment, a cost benefit analysis. *Australian Dental Journal*, 20, 316-325.
- Brown, L. M., Casamassimo, P. S., Griffen, A. & Tatakis, D. 2006. Supragingival calculus in children with gastrostomy feeding: significant reduction with a caregiver-applied tartar-control dentifrice. *Pediatric Dentistry* [Online], 28. Available: <http://onlinelibrary.wiley.com/o/cochrane/clcentral/articles/790/CN-00572790/frame.html>.

- Butcher, J. S., Wolraich, M. L., Gillaspy, S. R., Martin, V. G. & Wild, R. C. 2014. The impact of a Medical Home for children with developmental disability within a pediatric resident continuity clinic. *Journal of the Oklahoma State Medical Association*, 107, 632-638.
- Cardoso, A. M. R., Felipe, A. C. M., Nunes, F. M. R. & Padilha, W. W. N. 2012. Oral health home program for children and teenagers with cerebral palsy. *Pesquisa Brasileira em Odontopediatria e Clinica Integrada*, 12, 127-133.
- Carlstedt, K., Dahllof, G., Nilsson, B. & Modeer, T. 1996. Effect of palatal plate therapy in children with Down syndrome. A 1-year study. *Acta Odontologica Scandinavica*, 54, 122-125.
- Carrillo, C. M., Tanaka, M. H., Cesar, M. F., Camargo, M. A., Juliano, Y. & Novo, N. F. 2008. Use of papain gel in disabled patients. *Journal of Dentistry for Children (Chicago, Ill.)* [Online], 75. Available: <http://onlinelibrary.wiley.com/doi/10.1002/jdr.1253>
- Cermak, S. A., Stein Duker, L. I., Williams, M. E., Dawson, M. E., Lane, C. J. & Polido, J. C. 2015. Sensory adapted dental environments to enhance oral care for children with autism spectrum disorders: A randomized controlled pilot study. *Journal of Autism and Developmental Disorders*.
- Chang, J., Patton, L. L. & Kim, H. Y. 2014. Impact of dental treatment under general anesthesia on the oral health-related quality of life of adolescents and adults with special needs. *European Journal of Oral Sciences*, 122, 363-371.
- Charteris, P. & Kinsella, T. 2001. The Oral Care Link Nurse: a facilitator and educator for maintaining oral health for patients at the Royal Hospital for neuro-disability. *Special Care in Dentistry*, 21, 68-71.
- Chibinski, A. C., Pochapski, M. T., Farago, P. V., Santos, F. A. & Czlusniak, G. D. 2011. Clinical evaluation of chlorhexidine for the control of dental biofilm in children with special needs. *Community Dental Health*, 28, 222-226.
- Cildir, S. K., Sandalli, N., Nazli, S., Alp, F. & Caglar, E. 2012. A novel delivery system of probiotic drop and its effect on dental caries risk factors in cleft lip/palate children. *Cleft Palate Craniofacial Journal*, 49, 369-72.
- Cohen, S., Sarnat, H. & Shalgi, G. 1991. The role of instruction and a brushing device on the oral hygiene of blind children. *Clinical Preventive Dentistry*, 13, 8-12.
- Colodny, N., Miller, L. & Faralli, M. 2015. The development of a feeding, swallowing and oral care program using the PRECEDE-PROCEED model in an orphanage-hospital in Guatemala. *International Journal of Speech-Language Pathology*, 17, 127-137.
- De Abreu, M. H., Paixao, H. H., Resende, V. L. & Pordeus, I. A. 2002. Mechanical and chemical home plaque control: a study of Brazilian children and adolescents with disabilities. *Special Care in Dentistry*, 22, 59-64.
- De Andrade Meyer, A. C., De Mello Tera, T., Da Rocha, J. C. & Jardini, M. a. N. 2010. Clinical and microbiological evaluation of the use of toothpaste containing 1% chlorhexidine and the influence of motivation on oral hygiene in patients with motor deficiency. *Special Care in Dentistry*, 30, 140-145.
- De Oliveira Guaré, R., Ciamponi, A. L. & Romano, M. M. 2008. Behavioral and physiological changes in children with Down syndrome using mechanical and chemomechanical (Carisolv™) caries removal methods. *Special Care in Dentistry*, 28, 195-200.
- DeMattei, R. R., Allen, J. & Goss, B. 2012. A Service-Learning Project to Eliminate Barriers to Oral Care for Children with Special Health Care Needs. *Journal of School Nursing*, 28, 168-174.
- Dias, G. G., Prado, E. F. G. B., Vadasz, E. & Siqueira, J. T. T. 2010. Evaluation of the efficacy of a dental plaque control program in autistic patients. *Journal of Autism and Developmental Disorders*, 40, 704-708.
- Doykos 3rd, J. D., Sweeney, E. A. & Glass, R. L. 1967. Oral hygiene maintenance in handicapped children. *Journal of Dentistry for Children*, 34, 511-514.

- Farman, A. G., Horsley, B., Warr, E., Ianke, J. L. & Hood, H. 2003. Outcomes of digital X-ray mini-panel examinations for patients having mental retardation and developmental disability. *Dentomaxillofacial Radiology*, 32, 15-20.
- Faulks, D. & Hennequin, M. 2000. Evaluation of a long-term oral health program by carers of children and adults with intellectual disabilities. *Special Care in Dentistry*, 20, 199-208.
- Ferraz Dos Santos, B., Dabbagh, B., Daniel, S. J. & Schwartz, S. 2015. Association of onabotulinum toxin A treatment with salivary pH and dental caries of neurologically impaired children with sialorrhea. *International Journal of Paediatric Dentistry*.
- Full, C. A., Kerber, P. E., Boender, P. & Schneberger, N. 1977. Oral health maintenance of the institutionalized handicapped child. *The Journal of the American Dental Association*, 94, 111-113.
- Fuller, L. & Dunn, M. J. 1966. An occupational therapist's role in oral hygiene for the handicapped. *American Journal of Occupational Therapy*, 20, 35-6.
- Gallagher, I. H. C. & Cutress, T. W. 1977. Clinical trial in mentally retarded of chlorhexidine dental gel: bacteriology. *Community Dentistry and Oral Epidemiology*, 5, 1-6.
- Geary, J. L., Kinirons, M. J., Boyd, D. & Gregg, T. A. 2000. Individualized mouth prop for dental professionals and carers. *International Journal of Paediatric Dentistry*, 10, 71-74.
- Gertenrich, R. L. & Hart, R. W. 1972. Utilization of the oral hygiene team in a mental health institution. *Journal of Dentistry for Children*, 39, 174-177.
- Gertenrich, R. L. & Lewis, M. J. 1967. A study of automatic and hand tooth brushing as used on retarded or handicapped patients. *Journal of Dentistry for Children* [Online], 34. Available: <http://onlinelibrary.wiley.com/o/cochrane/clcentral/articles/835/CN-00271835/frame.html>.
- Glassman, P. & Miller, C. 2009. Social supports and prevention strategies as adjuncts and alternatives to sedation and anesthesia for people with special needs. *Special Care in Dentistry*, 29, 31-38.
- Goyal, S., Thomas, B. S., Bhat, K. M. & Subraya Bhat, G. 2011. Manual toothbrushing reinforced with audiovisual instruction versus powered toothbrushing among institutionalized mentally challenged subjects-A randomized cross-over clinical trial. *Medicina Oral, Patologia Oral y Cirugia Bucal*, 16, 359-364.
- Graudins, M. M., Rehfeldt, R. A., Demattei, R., Baker, J. C. & Scaglia, F. 2012. Exploring the efficacy of behavioral skills training to teach basic behavior analytic techniques to oral care providers. *Research in Autism Spectrum Disorders*, 6, 978-987.
- Hebbal, M. & Ankola, A. V. 2012. Development of a new technique (ATP) for training visually impaired children in oral hygiene maintenance. *European Archives of Paediatric Dentistry*, 13, 244-247.
- Hobdell, M. H., Burt, B. A. & Longhurst, P. 1975. A method of planning a dental treatment program for an institutionalized population. *Community Dentistry and Oral Epidemiology*, 3, 166-173.
- Hohoff, A. & Ehmer, U. 1999. Short-term and long-term results after early treatment with the Castillo Morales stimulating plate. A longitudinal study. *Journal of Orofacial Orthopedics*, 60, 2-12.
- Holcomb, F. H., Taylor, P. P. & Saunders, W. A. 1970. Comparison of two oral hygiene devices for the physically handicapped. *ASDC Journal of Dentistry for Children*, 37, 325-9.
- Holland, T. J. & O'Mullane, D. M. 1990. The organisation of dental care for groups of mentally handicapped persons. *Community Dental Health*, 7, 285-293.
- Hopper, S. M., Babl, F. E., McCarthy, M., Tancharoen, C., Lee, K. J. & Oakley, E. 2011. A double blind, randomised placebo-controlled trial of topical 2%

- viscous lidocaine in improving oral intake in children with painful infectious mouth conditions. *BMC Pediatrics*, 11, 106.
- Hoyer, H. & Limbrock, G. J. 1990. Orofacial regulation therapy in children with Down syndrome, using the methods and appliances of Castillo-Morales. *Journal of Dentistry for Children*, 57, 442-444.
- Ibricevic, H. H., S.; Honkala, E.; Al-Quraini, W. 2005. A field trial on semi-annual fluoride varnish applications among the special needs schoolchildren. *Journal of Clinical Pediatric Dentistry*, 30, 135-138.
- Kass, L. 1979. Dental health program for the institutionally mentally retarded. *Dental Hygiene (Chicago)*, 53, 76-8.
- Kumar, S., Konde, S., Raj, S. & Agarwal, M. 2012. Effect of oral health education and fluoridated dentifrices on the oral health status of visually impaired children. *Contemporary Clinical Dentistry*, 3, 398-401.
- Lange, B. 1985. Effects of modelling on the oral health care of persons with mentally handicapping conditions. *Special Care in Dentistry*, 5, 255-260.
- Liu, H. Y., Hung, H. C., Hsiao, S. Y., Chen, H. S., Yen, Y. Y., Huang, S. T., et al. 2013. Impact of 24-month fluoride tablet program on children with disabilities in a non-fluoridated country. *Research in Developmental Disabilities*, 34, 2598-2605.
- Luoma, H., Nyman, A., Toivonen, A., Soderholm, S., Nuuja, T., Kantero, R. L., et al. 1979. Effect of caries in mentally handicapped children of addition of fluoride and bicarbonate-phosphate to dietary sugar products. *Scandinavian Journal of Dental Research*, 87, 197-207.
- Mabry, C. C. & Mosca, N. G. 2006. Interprofessional educational partnerships in school health for children with special oral health needs. *Journal of Dental Education*, 70, 844-850.
- MacKenzie, B., Halliday, M. S., Holloway, P. J. & Worthington, H. V. 1988. An investigation into some factors influencing the communication of health education messages for adolescent 'slow learners.'. *Health Education Research*, 3, 153-159.
- Mann, J., Wolnerman, J. S., Lavie, G., Carlin, Y., Meir, S. & Garfunkel, A. A. 1986. The effect of dental education and dental treatment on the dental status of a handicapped population: a longitudinal study. *Special Care in Dentistry*, 6, 180-181.
- Merrick, J. S., J. 2000. Preventive dental health for persons with Down syndrome. *International Journal of Adolescent Medicine and Health*, 12, 81-84.
- Molina, G. F., Faulks, D., Mazzola, I., Mulder, J. & Frencken, J. E. 2014. One-year survival of ART and conventional restorations in patients with disability. *BMC Oral Health* [Online], 14. Available: <http://onlinelibrary.wiley.com/doi/10.1186/s12958-014-0148-7>
- Morales-Chavez, M. C. & Nualart-Grollmus, Z. C. 2014. Retention of a resin-based sealant and a glass ionomer used as a fissure sealant in children with special needs. *Journal of Clinical Experimental Dentistry*, 6, e551-5.
- Mouradian, W. E. & Corbin, S. B. 2003. Addressing health disparities through dental-medical collaborations, part II. Cross-cutting themes in the care of special populations. *Journal of Dental Education*, 67, 1320-1326.
- Nandini, N. S. 2003. New insights into improving the oral health of visually impaired children. *Journal of the Indian Society of Pedodontic Preventive Dentistry*, 21, 142-3.
- O'Donnell, D. & Crosswaite, M. A. 1988. Dental health education for mentally handicapped children. *Journal of the Royal Society of Health*, 108, 8-10.
- O'Donnell, D. & Crosswaite, M. A. 1990. Dental health education for the visually impaired child. *Journal of the Royal Society of Health*, 110, 60.
- Ohito, F. A., Opinya, G. N. & Wang'ombe, J. 1993. Dental caries, gingivitis and dental plaque in handicapped children in Nairobi, Kenya. *East African Medical Journal*, 70, 71-4.

- Orellana, L. M., Martínez-Sanchis, S. & Silvestre, F. J. 2014. Training adults and children with an autism spectrum disorder to be compliant with a clinical dental assessment using a TEACCH-based approach. *Journal of Autism and Developmental Disorders*, 44, 776-785.
- Palin-Palokas, T., Nordblad, A. & Remes-Lyly, T. 1997. Video as a medium of oral health education for children with mental handicaps. *Special Care in Dentistry*, 17, 211-214.
- Patil, Y. B., Hegde-Shetiya, S., Kakodkar, P. V. & Shirahatti, R. 2011. Evaluation of a preventive program based on caries risk among mentally challenged children using the Cariogram model. *Community Dental Health*, 28, 286-291.
- Pilebro, C. & Bäckman, B. 2005. Teaching oral hygiene to children with autism. *International Journal of Paediatric Dentistry*, 15, 1-9.
- Reynolds, W. E. & Block, R. M. 1974. Evaluating the effectiveness of instruction in oral hygiene for mentally retarded boys. *Journal of Public Health Dentistry*, 34, 8-12.
- Richardson, B. A., Smith, D. C. & Hargreaves, J. A. 1977. Study of a fissure sealant in mentally retarded Canadian children. *Community Dentistry and Oral Epidemiology* [Online], 5. Available: <http://onlinelibrary.wiley.com/o/cochrane/clcentral/articles/335/CN-00420335/frame.html>.
- Richardson, B. A., Smith, D. C. & Hargreaves, J. A. 1981. A 5-year clinical evaluation of the effectiveness of a fissure sealant in mentally retarded Canadian children. *Community Dentistry and Oral Epidemiology* [Online], 9. Available: <http://onlinelibrary.wiley.com/o/cochrane/clcentral/articles/334/CN-00420334/frame.html>.
- Sandeep, V., Vinay, C., Madhuri, V., Rao, V., Uloopi, K. & Sekhar, R. 2014. Impact of visual instruction on oral hygiene status of children with hearing impairment. *Journal of Indian Society of Pedodontics and Preventive Dentistry*, 32, 39-43.
- Sanders, C., Kleinert, H. L., Boyd, S. E., Herren, C., Theiss, L. & Mink, J. 2008. Virtual patient instruction for dental students: Can it improve dental care access for persons with special needs? *Special Care in Dentistry*, 28, 205-213.
- Santos, M. T. & Manzano, F. S. 2007. Assistive stabilization based on the neurodevelopmental treatment approach for dental care in individuals with cerebral palsy. *Quintessence Int*, 38, 681-7.
- Schnuth, M. L. 1977. Dental health education for the blind. *Dental Hygiene (Chicago)*, 51, 499-501.
- Schwartz, B. H., Albino, J. E. & Bissell, G. D. 1978. Development and evaluation of a dental health training program for severely retarded children. *Journal of Dentistry for the Handicapped*, 4, 17-22.
- Shapira, J. 1996. A comprehensive 30-month preventive dental health program in a pre-adolescent population with Down's Syndrome: A longitudinal study. *Special Care in Dentistry*, 16, 33-37.
- Sharma, A., Arora, R., Kenchappa, M., Bhayya, D. P. & Singh, D. 2012. Clinical evaluation of the plaque-removing ability of four different toothbrushes in visually impaired children. *Oral Health & Preventive Dentistry* [Online], 10. Available: <http://onlinelibrary.wiley.com/o/cochrane/clcentral/articles/245/CN-00871245/frame.html>.
- Shaw, L., Harris, B. M., MacLaurin, E. T. & Foster, T. D. 1983. Oral hygiene in handicapped children: a comparison of effectiveness in the unaided use of manual and electric toothbrushes. *Dental Health*, 22, 4-5.
- Shetty, V., Hegde, A. M. & Varghese, E. 2013. A Novel Music based Tooth Brushing System for Blind Children. *Journal of Clinical Pediatric Dentistry*, 37, 251-255.
- Shetty, V., Kumar, J. & Hegde, A. 2014. Breaking the sound barrier: oral health education for children with hearing impairment. *Special Care in Dentistry*, 34, 131-7.

- Shyama, M., Al-Mutawa, S. A., Honkala, S. & Honkala, E. 2003. Supervised toothbrushing and oral health education program in Kuwait for children and young adults with Down syndrome. *Special Care in Dentistry*, 23, 94-99.
- Shyama, M., Honkala, E., Honkala, S. & Al-Mutawa, S. A. 2006. Effect of xylitol candies on plaque and gingival indices in physically disabled school pupils. *Journal of Clinical Dentistry* [Online], 17. Available: <http://onlinelibrary.wiley.com/o/cochrane/clcentral/articles/514/CN-00566514/frame.html>.
- Stabholz, A., Shapira, J., Shur, D., Friedman, M., Guberman, R. & Sela, M. N. 1991. Local application of sustained-release delivery system of chlorhexidine in Down's syndrome population. *Clinical Preventive Dentistry*, 13, 9-14.
- Steelman, R., Holmes, D. & Hamilton, M. 1996. Chlorhexidine spray effects on plaque accumulation in developmentally disabled patients. *Journal of Clinical Pediatric Dentistry*, 20, 333-336.
- Stefanovska, E., Nakova, M., Radojkova-Nikolovska, V. & Ristoska, S. 2010. Tooth-brushing intervention programme among children with mental handicap. *Bratisl Lek Listy*, 111, 299-302.
- Stephens, A. J., Sapsford, D. J. & Curzon, M. E. 1993. Intravenous sedation for handicapped dental patients: a clinical trial of midazolam and propofol. *British Dental Journal* [Online], 175. Available: <http://onlinelibrary.wiley.com/o/cochrane/clcentral/articles/589/CN-00094589/frame.html>.
- Svatun, B. & Gjermo, P. 1978. Oral hygiene, periodontal health and need for periodontal treatment among institutionalized mentally subnormal persons in Norway. *Acta Odontologica Scandinavica*, 36, 89-95.
- Teitelbaum, A. P., Pochapski, M. T., Jansen, J. L., Sabbagh-Haddad, A., Santos, F. A. & Czlusniak, G. D. 2009. Evaluation of the mechanical and chemical control of dental biofilm in patients with Down syndrome. *Community Dentistry and Oral Epidemiology*, 37, 463-467.
- Willette, J. C. & Savage, J. A. 1978. Positive motivation: A method for promoting oral health among mentally retarded people. *Mental Retardation*, 16, 233-235.
- Williams, N. J. & Schuman, N. J. 1988. The curved-bristle toothbrush: an aid for the handicapped population. *Journal of Dentistry for Children*, 55, 291-293.
- Winstanley, M. L. 1983. A synopsis of the project to evaluate the use of a Braille text and tactile aids when teaching dental health to blind children. *British Dental Surgery Assistant*, 42, 20-3.
- Wu, K. P.-H., Ke, J.-Y., Chen, C.-Y., Chen, C.-L., Chou, M.-Y. & Pei, Y.-C. 2011. Botulinum toxin type A on oral health in treating sialorrhea in children with cerebral palsy: A randomized, double-blind, placebo-controlled study. *Journal of Child Neurology*, 26, 838-843.
- Yalcinkaya, S. E. & Atalay, T. 2006. Improvement of oral health knowledge in a group of visually impaired students. *Oral Health & Preventive Dentistry*, 4, 243-253.
- Yoshihara, T., Morinushi, T., Kinjyo, S. & Yamasaki, Y. 2005. Effect of periodic preventive care on the progression of periodontal disease in young adults with Down's syndrome. *Journal of Clinical Periodontology*, 32, 556-560.
- Young, W. O. & Shannon, J. H. 1968. Providing dental treatment for handicapped children. *ASDC Journal of Dentistry for Children*, 35, 225-40.
- Zaldivar-Chiapa, R. M., Arce-Mendoza, A. Y., De La Rosa-Ramírez, M., Caffesse, R. G. & Solís-Soto, J. M. 2005. Evaluation of surgical and non-surgical periodontal therapies, and immunological status, of Young Down's syndrome patients. *Journal of Periodontology*, 76, 1061-1065.
- Zavaglia, V., Nori, A. & Mansour, N. M. 2003. Long term effects of the palatal plate therapy for the orofacial regulation in children with Down syndrome. *The Journal of Clinical Pediatric Dentistry*, 28, 89-93.

7.3 Cochrane Review: Included Studies

- Abramson E. E. & Wunderlich R. A. 1972. Dental hygiene training for retardates: an application of behavioral techniques. *Mental Retardation*, 10, 6-8.
- Albino, J. E., Schwartz, B. H., Goldberg, H. J. & Stern, M. E. 1979. Results of an oral hygiene program for severely retarded children. *ASDC Journal of Dentistry for Children*, 46, 25-28.
- Altabet, S., Rogers, K., Imes, E., Boatman Ida, M. & Moncier, J. 2003. Comprehensive Approach Toward Improving Oral Hygiene at a State Residential Facility for People with Mental Retardation. *Mental Retardation*, 41, 440-445.
- Bickley, S. R., Shaw, L. & Shaw, M. J. 1990. Use and effect of clinical photographic records on the motivation and oral hygiene practices of a group of mentally handicapped adults. *Dental Health*, 29, 3-6.
- Bildt, M. M. & Ekhardt G J C. 2010. The effect of a three-headed toothbrush on oral health in patients with severe disabilities. *International Association of Disability and Oral Health*. Journal of Disability and Oral Health.
- Bouter H. P. & Smeets P. M. 1979. Teaching toothbrushing behaviour in severely retarded adults: systematic reduction of feedback and duration training. *International Journal of Rehabilitation Research*, 2, 61-69.
- Bratel, J. & Berggren, U. 1991. Long-term oral effects of manual or electric toothbrushes used by mentally handicapped adults. *Clinical Preventive Dentistry*, 13, 5-7.
- Bratel, J., Berggren, U. & Hirsch, J. M. 1988. Electric or manual toothbrush? A comparison of the effects on the oral health of mentally handicapped adults. *Clinical Preventive Dentistry*, 10, 23-26.
- Carr, M. P., Sterling, E. S. & Bauchmoyer, S. M. 1997. Comparison of the Interplak and manual toothbrushes in a population with mental retardation/developmental disabilities (MR/DD). *Special Care in Dentistry*, 17, 133-136.
- Christen, S. 2007. *Klinische Untersuchung der Putzeffektivität und Handhabbarkeit einer elektrischen Dreikopfbürste, einer dreiköpfigen Handzahnbürste sowie einer konventionellen Handzahnbürste im Vergleich bei Patienten mit Behinderungen / A clinical investigation of a comparison between the brushing effectiveness and manageability of a three-head electric toothbrush, a three-headed manual toothbrush and a conventional manual toothbrush in patients with disabilities*. Dr. med. Dent., The University Medicine Berlin.
- Dogan, M. C., Alacam, A., Asici, N., Odabas, M. & Seydaoglu, G. 2004. Clinical evaluation of the plaque-removing ability of three different toothbrushes in a mentally disabled group. *Acta Odontologica Scandinavica*, 62, 350-354.
- Ferozali, F., Johnson, G. & Cavagnaro, A. 2007. Health benefits and reductions in bacteria from enhanced oral care. *Special Care in Dentistry*, 27, 168-176.
- Garcia-Carrillo, A., Jover, A., Pla, R., Martorell, A., Sota, C., Gomez-Moreno, G., et al. 2016. Manual versus sonic powered toothbrushing in patients with intellectual disability: a cluster-randomized clinical trial. *Journal of Clinical Periodontology*, 43, 684-693.
- Gertenrich, R. L. & Lewis, M. J. 1967. A study of automatic and hand tooth brushing as used on retarded or handicapped patients. *Journal of Dentistry for Children*, 34, 145-164.
- Glassman, P. & Miller, C. E. 2006. Effect of preventive dentistry training program for caregivers in community facilities on caregiver and client behavior and client oral hygiene. *New York State Dental Journal*, 72, 38-46.
- Gonzalez, E. E., Nathe, C. N., Logothetis, D. D., Pizanis, V. G. & Sanchez-Dils, E. 2013. Training caregivers: disabilities and dental hygiene. *International Journal of Dental Hygiene*, 11, 293-297.
- Herrera, D. 2015. Manual versus sonic powered tooth brushing in patients with intellectual disability. 24th August 2015 ed. Spain: ISRCTN Registry.

- Jarman P. H., Iwata B. A. & Lorentzson A. M. 1983. Development of morning self-care routines in multiply handicapped persons. *Applied Research in Mental Retardation*, 4, 113-122.
- Kaschke, I. 2008. *Mundgesundheitsförderung für erwachsene Bewohner von Behinderteneinrichtungen – Evaluation des Berliner Modellprojektes / Oral health promotion for adult residents of disabled facilities – evaluation of the Berlin Model project*. Masters Thesis, Freie Universität Berlin.
- Kaschke, I., Klaus -Roland, J. & Zeller, A. 2005. The effectiveness of different toothbrushes for people with special needs. *Journal of Disability and Oral Health*, 6, 65-71.
- Kelner, M. 1963. Comparative Analysis of the Effect of Automatic and Conventional Toothbrushing in Mental Retardates. *Pennsylvania Dental Journal*, 30, 102 - 109.
- Kissel R. C., Whitman T. L. & Reid D, H. 1983. An institutional staff training and self-management program for developing multiple self-care skills in severely/profoundly retarded individuals. *Journal of Applied Behavior Analysis*, 16, 395-415.
- Lange, B. 1985. Effects of modelling on the oral health care of persons with mentally handicapping conditions. *Special Care in Dentistry*, 5, 255-260.
- Lange, B., Cook, C., Dunning, D., Froeschle, M. L. & Kent, D. 2000. Improving the oral hygiene of institutionalized mentally retarded clients. *Journal of Dental Hygiene*, 74, 205-209.
- Lange, B. M. 1981. Effects of modeling on the oral health care of mentally retarded. *Dissertation Abstracts International*, 41, 4654.
- Mac Giolla Phadraig, C., Guerin, S. & Nunn, J. 2013. Train the trainer? A randomized controlled trial of a multi-tiered oral health education programme in community-based residential services for adults with intellectual disability. *Community Dental and Oral Epidemiology* 41, 182-92.
- Mac Giolla Phadraig, C., Guerin, S. & Nunn, J. 2013. Train the trainer? A randomized controlled trial of a multi-tiered oral health education programme in community-based residential services for adults with intellectual disability. *Community Dentistry and Oral Epidemiology*, 41, 182-192.
- Mac Giolla Phadraig, C., Guerin, S. & Nunn, J. 2015. Should we educate care staff to improve the oral health and oral hygiene of people with intellectual disability in residential care? Real world lessons from a randomized controlled trial. *Special Care in Dentistry*, 35, 92-98.
- Olmos, I. 2015. *Mundgesundheitsförderung bei erwachsenen Bewohnern mit Behinderung in Behinderteneinrichtungen – Evaluation der Nachhaltigkeit des Berliner Modellprojektes/ Oral health promotion in adult handicapped persons in disabled people's facilities – evaluation of the sustainability of the Berlin model project*. Masters Thesis, Freie Universität Berlin.
- Olmos, I. & Kaschke, I. 2016. Evaluation des Berliner Mundgesundheitsprogramms für erwachsene Bewohner von Behinderteneinrichtungen / Evaluation of the Berlin Oral health programme for adult residents of disabled facilities. *Prophylaxe Impuls / Prophylaxis Impulse*, 20. Jahrgang / 20th Year, 6 - 13.
- Phlypo, I. 2016. Integrating community service-learning in undergraduate dental education: an oral health project in a residential facility for people with intellectual disabilities. Unpublished study.
- Phlypo, I. 2016. Pilot Study on the Effects of Oral Health Education for Caregivers in a Residential Setting for People with Intellectual Disabilities: A Randomized Controlled Trial. *International Association of Disability and Oral Health* Chicago: Special Care Dentistry.
- Sauvetre, E., Rozow, A., De Meel, H., Richebe, A., Abi-Khalil, M. & Demeure, F. 1995. Comparison of the clinical effectiveness of a single and a triple-headed toothbrush in a population of mentally retarded patients. *Bulletin du Groupement International pour la Recherche Scientifique en Stomatologie & Odontologie*, 38, 115-119.

- Schwartz, B. H., Albino, J. E. & Bissell, G. D. 1978. Development and evaluation of a dental health training program for severely retarded children. *Journal of Dentistry for the Handicapped*, 4, 17-22.
- Shaw, L., Harris, B. M., MacLaurin, E. T. & Foster, T. D. 1983. Oral hygiene in handicapped children: a comparison of effectiveness in the unaided use of manual and electric toothbrushes. *Dental Health*, 22, 4-5.
- Shaw, L. & Shaw, M. J. 1990. The effectiveness of differing dental health education programmes in improving the oral health of adults with mental handicaps. *Journal of Dental Research*, 69, 973.
- Shaw, M. J. & Shaw, L. 1991. The effectiveness of differing dental health education programmes in improving the oral health of adults with mental handicaps attending Birmingham adult training centres. *Community Dental Health*, 8, 139-145.
- Shaw, M. J., Shaw, L. & Foster, T. D. 1990. The oral health in different groups of adults with mental handicaps attending Birmingham (UK) adult training centres. *Community Dental Health*, 7, 135-141.
- Snell M. E., Lewis, A. P. & Houghton, A. 1989. Acquisition and maintenance of toothbrushing skills by students with cerebral palsy and mental retardation. *Journal of the Association for Persons with Severe Handicaps*, 14, 216-226.
- Swallow, J. N., Davies, D. E. & Hawkins, S. D. 1969. Gingival disease prevalence in mentally handicapped adults. The effects of an oral hygiene programme. *British Dental Journal*, 127, 376-379.
- Teitelbaum, A. P., Pochapski, M. T., Jansen, J. L., Sabbagh-Haddad, A., Santos, F. A. & Czlusniak, G. D. 2009. Evaluation of the mechanical and chemical control of dental biofilm in patients with Down syndrome. *Community Dentistry and Oral Epidemiology*, 37, 463-467.
- Williams, N. J. & Schuman, N. J. 1988. The curved-bristle toothbrush: an aid for the handicapped population. *Journal of Dentistry for Children*, 55, 291-293.
- Zeller, A. 2004. *Klinische Untersuchung der Putzeffektivität und Handhabbarkeit einer elektronischen Zahnbürste, einer dreiköpfigen Handzahnbürste und einer konventionellen Handzahnbürste bei Patienten mit Behinderungen / Clinical investigation of the brushing effectiveness and manageability of an electronic toothbrush, a three-headed manual toothbrush and a conventional manual toothbrush in patients with disabilities.*, Freie Universität Berlin

7.4 Realist Review: Included Studies

- Adiwoso, A. S. & Pilot, T. 1999. Results of oral health and hygiene education in an institution for multiple handicapped children in Indonesia. *International Dental Journal*, 49, 82-9.
- Albrecht, M., Kupfer, R., Reissmann, D. R., Mühlhauser, I. & Köpke, S. 2016. Oral health educational interventions for nursing home staff and residents. *Cochrane Database of Systematic Reviews*.
- Altabet, S., Rogers, K., Imes, E., Boatman Ida, M. & Moncier, J. 2003. Comprehensive Approach Toward Improving Oral Hygiene at a State Residential Facility for People with Mental Retardation. *Mental Retardation*, 41, 440-445.
- Aylward, S., Stolee, P., Keat, N. & Johncox, V. 2003. Effectiveness of Continuing Education in Long-Term Care: A Literature Review. *The Gerontologist*, 43, 259-271.
- Barnes, M., Henwood, F. & Smith, N. 2016. Information and care: A relational approach. *Dementia (London)*, 15, 510-25.
- Binkley, C. J., Johnson, K. W., Abadi, M., Thompson, K., Shamblen, S. R., Young, L., et al. 2014. Improving the oral health of residents with intellectual and developmental disabilities: an oral health strategy and pilot study. *Evaluation and Program Planning*, 47, 54-63.

- Blinkhorn, F. A., Weingarten, L., Boivin, L., Plain, J. & Kay, M. 2012. An intervention to improve the oral health of residents in an aged care facility led by nurses. *Health Education Journal*, 71, 527-535.
- Brady, M., Furlanetto, D., Hunter, R. V., Lewis, S. & Milne, V. 2006. Staff-led interventions for improving oral hygiene in patients following stroke. *Cochrane Database Systematic Reviews*, Cd003864.
- Chadwick, D., Chapman, M. & Davies, G. 2017. Factors affecting access to daily oral and dental care among adults with intellectual disabilities. *Journal of applied research in intellectual disabilities*, 31, 379-394.
- Chiba, Y., Shimoyama, K. & Suzuki, Y. 2009. Recognition and behaviour of caregiver managers related to oral care in the community. *Gerodontology*, 26, 112-121.
- Coker, E., Ploeg, J. & Kaasalainen, S. 2014. The Effect of Programs to Improve Oral Hygiene Outcomes for Older Residents in Long-Term Care A Systematic Review. *Research in Gerontological Nursing*, 7, 87-100.
- Colodny, N., Miller, L. & Faralli, M. 2015. The development of a feeding, swallowing and oral care program using the PRECEDE-PROCEED model in an orphanage-hospital in Guatemala. *International Journal of Speech-Language Pathology*, 17, 127-137.
- Crawford, H., Leslie, P. & Drinnan, M. J. 2007. Compliance with dysphagia recommendations by carers of adults with intellectual impairment. *Dysphagia*, 22, 326-34.
- Cullen-Erickson, M. 1994. Parents' experiences in assisting a person with an intellectual disability to achieve optimal dental health. *Australian Occupational Therapy Journal*, 41, 163-172.
- Davies, K. W. & Whittle, J. G. 1990. Dental health education: training of homecarers of mentally handicapped adults. *Community Dent Health*, 7, 193-7.
- De Lugt-Lustig, K. H. M. E., Vanobbergen, J. N. O., Van Der Putten, G.-J., De Visschere, L. M. J., Schols, J. M. G. A. & De Baat, C. 2014. Effect of oral healthcare education on knowledge, attitude and skills of care home nurses: a systematic literature review. *Community Dentistry and Oral Epidemiology*, 42, 88-96.
- De Visschere, L., De Baat, C., Schols, J. & Deschepper, E., Vanobbergen, J., 2011. Evaluation of the implementation of an 'oral hygiene protocol' in nursing homes: a 5-year longitudinal study. *Community Dentistry and Oral Epidemiology*, 39, 416-425.
- De Visschere, L., Schols, J., Van Der Putten, G. J., De Baat, C. & Vanobbergen, J. 2012. Effect evaluation of a supervised versus non-supervised implementation of an oral health care guideline in nursing homes: a cluster randomised controlled clinical trial. *Gerodontology*, 29, E96-E106.
- Duane B., Kirk D. & Forbes G. M., Ball G. 2011. Inspiring and recognising good oral health practice within care homes. *Journal of Disability and Oral Health*, 12, 3-9.
- Eijssink, A. M., Schipper, G. & Vermaire, J. H. 2018. A Q-methodology study among caregivers of people with moderate intellectual disabilities on their clients' health care: An example in oral health. *Journal of Applied Research in Intellectual Disabilities*, 0.
- Faulks, D. & Hennequin, M. 2000. Evaluation of a long-term oral health program by carers of children and adults with intellectual disabilities. *Special Care in Dentistry*, 20, 199-208.
- Fickert N. A. & Ross, D. 2012. Effectiveness of a caregiver education program on providing oral care to individuals with intellectual and developmental disabilities. *Intellectual and developmental disabilities*, 50, 219-232.
- Forsell, M. K., E.; Hoogstraate, J.; Herbst, B.; Johansson, O.; Sjogren, P. 2010. A Survey of Attitudes and Perceptions Toward Oral Hygiene Among Staff at a Geriatric Nursing Home. *Geriatric Nursing*, 31, 435-440.

- Forsell, M. S., P.; Kullberg, E.; Johansson, O.; Wedel, P.; Herbst, B.; Hoogstraate, J. 2011. Attitudes and perceptions towards oral hygiene tasks among geriatric nursing home staff. *International Journal of Dental Hygiene*, 9, 199-203.
- Gammack, J. K. & Pulisetty, S. 2009. Nursing Education and Improvement in Oral Care Delivery in Long-Term Care. *Journal of the American Medical Directors Association*, 10, 658-661.
- Glassman, P. & Miller, C. E. 2006. Effect of preventive dentistry training program for caregivers in community facilities on caregiver and client behavior and client oral hygiene. *New York State Dental Journal*, 72, 38-46.
- Goh, C. E., Guay, M. P., Lim, M. Y., Lim, S. M., Loke, S. Y., Toh, H. E., et al. 2016. Correlates of attitudes and perceived behavioural control towards oral care provision among trained and untrained nursing home caregivers in Singapore. *Journal of Clinical Nursing*, 25, 1624-1633.
- Gonzalez, E. E., Nathe, C. N., Logothetis, D. D., Pizanis, V. G. & Sanchez-Dils, E. 2013. Training caregivers: disabilities and dental hygiene. *International Journal of Dental Hygiene*, 11, 293-297.
- Hammar, L. M., Swall, A. & Meranius, M. S. 2016. Ethical aspects of caregivers' experience with persons with dementia at mealtimes. *Nursing Ethics*, 23, 624-35.
- Hithersay, R., Strydom, A., Moulster, G. & Buszewicz M. 2014. Carer-led health interventions to monitor, promote and improve the health of adults with intellectual disabilities in the community: A systematic review. *Research in Developmental Disabilities*, 35, 887-907.
- Jablonski-Jaudon, R. A., Kolanowski, A. M., Winstead, V., Jones-Townsend, C. & Azuero, A. 2016. Maturation of the MOUTH Intervention: from Reducing Threat to Relationship-Centered Care. *Journal of Gerontological Nursing*, 42, 15-23; quiz 24-5.
- Janssens, B., De Visschere, L., Van Der Putten, G. J., De Lugt-Lustig, K. & Schols, J. V., J. 2016. Effect of an oral healthcare protocol in nursing homes on care staffs' knowledge and attitude towards oral health care: a cluster-randomised controlled trial. *Gerodontology*, 33, 275-286.
- Janssens, B., Vanobbergen, J., Lambert, M., Schols, J. & De Visschere, L. 2018. Effect of an oral healthcare programme on care staff knowledge and attitude regarding oral health: a non-randomised intervention trial. *Clinical Oral Investigations*, 22, 281-292.
- Kissel R. C., Whitman T., L. & Reid D., H. 1983. An institutional staff training and self-management program for developing multiple self-care skills in severely/profoundly retarded individuals. *Journal of Applied Behavior Analysis*, 16, 395-415.
- Kohli, R., Nelson, S., Ulrich, S., Finch, T., Hall, K. & Schwarz, E. 2017. Dental care practices and oral health training for professional caregivers in long-term care facilities: An interdisciplinary approach to address oral health disparities. *Geriatric Nursing*, 38, 296-301.
- Kullberg, E., Forsell, M., Wedel, P., Sjoogren, P., Johansson, O., Herbst, B., et al. 2009. Dental Hygiene Education for Nursing Staff. *Geriatric Nursing*, 30, 329-333.
- Kullberg, E., Sjoogren, P., Forsell, M., Hoogstraate, J., Herbst, B. & Johansson, O. 2010. Dental hygiene education for nursing staff in a nursing home for older people. *Journal of Advanced Nursing*, 66, 1273-1279.
- Lange, B., Cook, C., Dunning, D., Froeschle, M. L. & Kent, D. 2000. Improving the oral hygiene of institutionalized mentally retarded clients. *Journal of Dental Hygiene*, 74, 205-209.
- Mac Giolla Phadraig, C., Guerin, S. & Nunn, J. 2013. Train the trainer? A randomized controlled trial of a multi-tiered oral health education programme in community-based residential services for adults with intellectual disability. *Community Dental and Oral Epidemiology* 41, 182-92.

- Mac Giolla Phadraig, C., Guerin, S. & Nunn, J. 2015. Should we educate care staff to improve the oral health and oral hygiene of people with intellectual disability in residential care? Real world lessons from a randomized controlled trial. *Special Care in Dentistry*, 35, 92-8.
- Mac Giolla Phadraig, C., Nunn, J., Guerin, S. & Normand, C. 2016. Should we provide oral health training for staff caring for people with intellectual disabilities in community based residential care? A cost-effectiveness analysis. *Evaluation and program planning*, 55, 46-54.
- Minihan, P. M., Morgan, J. P., Park, A., Yantsides, K. E., Nobles, C. J., Finkelman, M. D., et al. 2014. At-home oral care for adults with developmental disabilities: a survey of caregivers. *Journal of the American Dental Association*, 145, 1018-25.
- Nicol, R., Sweeney, M. P., McHugh, S. & Bagg, J. 2005. Effectiveness of health care worker training on the oral health of elderly residents of nursing homes. *Community Dentistry and Oral Epidemiology*, 33, 115-124.
- Nicolaci, A. B. & Tesini, D. A. 1982. Improvement in the oral hygiene of institutionalized mentally retarded individuals through training of direct care staff: a longitudinal study. *Special Care in Dentistry*, 2, 217-221.
- Pradhan, A., Keuskamp, D. & Brennan, D. 2016. Pre-and post-training evaluation of dental efficacy and activation measures in carers of adults with disabilities in South Australia—a pilot study. *Health & Social Care in the Community*, 24, 739-746.
- Risma, K. M., Weber-Gasparoni, K., Swenson, S. E., Ettinger, R. L. & Qian, F. 2015. Group home caregivers' comfort levels regarding physical resistance during oral hygiene care. *Special Care in Dentistry*, 35, 123-31.
- Shah, A. H., Naseem, M., Khan, M. S., Asiri, F. Y. I., Alqarni, I., Gulzar, S., et al. 2017. Oral health knowledge and attitude among caregivers of special needs patients at a Comprehensive Rehabilitation Centre: an analytical study. *Annali Di Stomatologia*, 8, 110-116.
- Shimoyama, K., Chiba, Y. & Suzuki, Y. 2007. The effect of awareness on the outcome of oral health performed by home care service providers. *Gerodontology*, 24, 204-210.
- Simons, D., Baker, P., Jones, B., Kidd, E. & Beighton, D. 2000. Dental health education: An evaluation of an oral health training programme for carers of the elderly in residential homes. *British Dental Journal*, 188, 206.
- Thole, K., Chalmers, J., Ettinger, R. L. & Warren, J. 2010. Iowa intermediate care facilities: An evaluation of care providers' attitudes toward oral hygiene care. *Special Care in Dentistry*, 30, 99-105.
- Van Der Putten, G. J., Mulder, J., De Baat, C., De Visschere, L. M. J., Vanobbergen, J. N. O., & Schols, J. 2013. Effectiveness of supervised implementation of an oral health care guideline in care homes; a single-blinded cluster randomized controlled trial. *Clinical Oral Investigations*, 17, 1143-1153.
- Wadsworth, C. L., Farrington, F. H., Schroeder, J. R. & Neel, N. L. 1986. The successes and failures of two approaches to dental care in institutions for patients who are mentally handicapped. *Special care in dentistry*, 6, 175-176.
- Wang, T. F., Huang, C. M., Chou, C. & Yu, S. 2015. Effect of oral health education programs for caregivers on oral hygiene of the elderly: A systemic review and meta-analysis. *International Journal of Nursing Studies*, 52, 1090-1096.
- Wardh, I., Berggren, U., Andersson, L. & Sorensen, S. 2002. Assessments of oral health care in dependent older persons in nursing facilities. *Acta Odontologica Scandinavica*, 60, 330-336.
- Wardh, I., Hallberg, L. R. M., Berggren, U., Andersson, L. & Sorensen, S. 2003. Oral health education for nursing personnel; experiences among specially trained oral care aides: One-year follow-up interviews with oral care aides at a nursing facility. *Scandinavian Journal of Caring Sciences*, 17, 250-256.
- Weening-Verbree L., Van Huisman-De Waal G., Van Dusseldorp L., Achterberg T. & Schoonhoven L. 2013. Oral health care in older people in long term care

- facilities: A systematic review of implementation strategies. *International Journal of Nursing Studies*, 50, 569-582.
- Willette, J. C. & Savage, J. A. 1978. Positive motivation: A method for promoting oral health among mentally retarded people. *Mental Retardation*, 16, 233-235.
- Zizzi, A., Piemontese, M., Gesuita, R., Nori, A., Berlin, R. S., Rocchetti, R., et al. 2014. Periodontal status in the Down's syndrome subjects living in central-eastern Italy: the effects of place of living. *International Journal of Dental Hygiene*, 12, 193-198.