Appendix 17: Medical Chart and Structured Interview Record Sheet

Medical Chart & Structured Interview Record Sheet
#STUDY ID _____

note: parent and child/teen present; encourage child/teen to answer independently where possible, parent to assist only when child/teen cannot answer

Diagnosis/es _____

Additional diagnosis/es _____

Medications _____

Presenting symptom/s at onset_____ _____

Any event prior to initial onset symptom/s?

(circle) Injury/Virus/Other _____

Anything that you associate with ongoing symptoms? _____

Multisystem complaints

Medical chart record		**Participant report**	
Skin	**Yes/No**	**Skin**	**Yes/No**
easy bruising/other _____		easy bruising/other_____	
Eyes	**Yes/No**	**Eyes**	**Yes/No**
dry eyes/other _____		dry eyes/other _____	
Cardiovascular	**Yes/No**	**Cardiovascular**	**Yes/No**
dizziness/fainting		dizziness/fainting	
other_____		other _____	
Gastrointestinal	**Yes/No**	**Gastrointestinal**	**Yes/No**
pain/constipation/diarrhoea		pain/constipation/diarrhoea	
other _____		other _____	
Urinary	**Yes/No**	**Urinary**	**Yes/No**
incontinence/bedwetting/other_____		incontinence/bedwetting/other_____	
Headache	**Yes/No**	**Headache**	**Yes/No**
Other_____		**Other**_____	

Medical Chart & Structured Interview Record Sheet
#STUDY ID _____

Diagnosis

Length of time to diagnosis (months) _____

Where did diagnosis occur Tertiary (type)_____/GP/other_____

Current schooling (circle) In- school home-school

Engagement with schooling as perceived by child/parent to be due to diagnosis

% of time _____

Chronically painful joints (**3 months or greater**) Number _____

Subluxed or dislocated joints as perceived in the past 3 months Yes/No

If no – skip to management

Number _____

A and E attendance past year (circle) Yes/No

Investigations past year (circle) XRAY/MRI/US/Other

Management past year (circle) Physio/Surgery/other (list) _____

comments_____

What is your ethnic group/background?	**X the box**
White	Irish ☐
	Irish Traveller ☐
	Roma ☐
	any other white background ☐
Black or Black Irish	African ☐
	Any other black background ☐
Asian or Asian Irish	Chinese ☐
	Indian/Pakistani/Bangladeshi ☐
	Any other Asian background ☐ Other
including mixed group, background	Other ☐
Prefer not to say	☐

Appendix 18: Clinical Data Collection Sheet

Clinical Data Collection Sheet
#STUDYID _____

1. 6MWT

Record the number of laps from the counter_____

Record the additional distance covered _____

(Number of meters in the final partial lap using the markers on the wall as distance guides)

Calculate the total distance walked (6MWD) rounding to the nearest meter

_____

Borg Breathlessness Scale **PRE** ☐ **POST** ☐

2. Y-Balance Test
Six practice trials; each leg; each direction; testing order

Standard testing order **three trials per reach**

Record to the nearest 0.5cm

Right Anterior	1.	2.	3.
Left Anterior	1.	2.	3.
Right Posteromedial	1.	2.	3.
Left Posteromedial	1.	2.	3.
Right posterolateral	1.	2.	3.
Left posterolateral	1.	2.	3.

3. Leg Length to the nearest 0.1cm _____

4. Height to the nearest 0.1cm _____

5. Weight to the nearest 0.1kg _____

6. LLAS see LLAS score sheet

7. Hamstring Length KEA degrees ☐

8. Leg Lift Endurance (straight leg lift/duration) No. of seconds ☐
(circle)
0 = Unable
I = 1-9seconds
2 = 10-29 seconds
3 = 30-59 seconds
4 = 60-119 seconds
5 = >2 minutes

9. Beighton see Beighton score sheet ☐ /9

10. FPI see FPI score sheet

11. Grip Strength

Record the scores of three hands trials for each hand tested.

Right hand	1.	2.	3.
Left hand	1.	2.	3.

12. BOT-2B see BOT-2B score sheet

Appendix 19: PedsQL Quality of Life Questionnaire (Parent and Child)

ID# _____

Date:_____

PedsQL™

Pediatric Quality of Life Inventory

Version 4.0

TEEN REPORT (ages 13-18)

DIRECTIONS

On the following page is a list of things that might be a problem for you. Please tell us **how much of a problem** each one has been for you during the **past ONE month** by circling:

0 if it is **never** a problem
1 if it is **almost never** a problem
2 if it is **sometimes** a problem
3 if it is **often** a problem
4 if it is **almost always** a problem

There are no right or wrong answers.
If you do not understand a question, please ask for help.

*In the past **ONE month**, how much of a **problem** has this been for you …*

About My Health and Activities *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard for me to walk more than one block	0	1	2	3	4
2. It is hard for me to run	0	1	2	3	4
3. It is hard for me to do sports activity or exercise	0	1	2	3	4
4. It is hard for me to lift something heavy	0	1	2	3	4
5. It is hard for me to take a bath or shower by myself	0	1	2	3	4
6. It is hard for me to do chores around the house	0	1	2	3	4
7. I hurt or ache	0	1	2	3	4
8. I have low energy	0	1	2	3	4

About My Feelings *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. I feel afraid or scared	0	1	2	3	4
2. I feel sad or blue	0	1	2	3	4
3. I feel angry	0	1	2	3	4
4. I have trouble sleeping	0	1	2	3	4
5. I worry about what will happen to me	0	1	2	3	4

How I Get Along with Others *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. I have trouble getting along with other teens	0	1	2	3	4
2. Other teens do not want to be my friend	0	1	2	3	4
3. Other teens tease me	0	1	2	3	4
4. I cannot do things that other teens my age can do	0	1	2	3	4
5. It is hard to keep up with my peers	0	1	2	3	4

About School *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard to pay attention in class	0	1	2	3	4
2. I forget things	0	1	2	3	4
3. I have trouble keeping up with my schoolwork	0	1	2	3	4
4. I miss school because of not feeling well	0	1	2	3	4
5. I miss school to go to the doctor or hospital	0	1	2	3	4

ID#_____

Date:_____

PedsQL™

Pediatric Quality of Life Inventory

Version 4.0

PARENT REPORT for TEENS (ages 13-18)

DIRECTIONS

On the following page is a list of things that might be a problem for **your teen**. Please tell us **how much of a problem** each one has been for **your teen** during the **past ONE month** by circling:

0 if it is **never** a problem
1 if it is **almost never** a problem
2 if it is **sometimes** a problem
3 if it is **often** a problem
4 if it is **almost always** a problem

There are no right or wrong answers.
If you do not understand a question, please ask for help.

*In the past **ONE month**, how much of a **problem** has your teen had with …*

PHYSICAL FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Walking more than one block	0	1	2	3	4
2. Running	0	1	2	3	4
3. Participating in sports activity or exercise	0	1	2	3	4
4. Lifting something heavy	0	1	2	3	4
5. Taking a bath or shower by him or herself	0	1	2	3	4
6. Doing chores around the house	0	1	2	3	4
7. Having hurts or aches	0	1	2	3	4
8. Low energy level	0	1	2	3	4

EMOTIONAL FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Feeling afraid or scared	0	1	2	3	4
2. Feeling sad or blue	0	1	2	3	4
3. Feeling angry	0	1	2	3	4
4. Trouble sleeping	0	1	2	3	4
5. Worrying about what will happen to him or her	0	1	2	3	4

SOCIAL FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Getting along with other teens	0	1	2	3	4
2. Other teens not wanting to be his or her friend	0	1	2	3	4
3. Getting teased by other teens	0	1	2	3	4
4. Not able to do things that other teens his or her age can do	0	1	2	3	4
5. Keeping up with other teens	0	1	2	3	4

SCHOOL FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Paying attention in class	0	1	2	3	4
2. Forgetting things	0	1	2	3	4
3. Keeping up with schoolwork	0	1	2	3	4
4. Missing school because of not feeling well	0	1	2	3	4
5. Missing school to go to the doctor or hospital	0	1	2	3	4

ID# _____

Date: _____

PedsQL™

Pediatric Quality of Life Inventory

Version 4.0

CHILD REPORT (ages 8-12)

DIRECTIONS

On the following page is a list of things that might be a problem for you. Please tell us **how much of a problem** each one has been for you during the **past ONE month** by circling:

0 if it is **never** a problem
1 if it is **almost never** a problem
2 if it is **sometimes** a problem
3 if it is **often** a problem
4 if it is **almost always** a problem

There are no right or wrong answers.
If you do not understand a question, please ask for help.

*In the past **ONE month**, how much of a **problem** has this been for you …*

About My Health and Activities *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard for me to walk more than one block	0	1	2	3	4
2. It is hard for me to run	0	1	2	3	4
3. It is hard for me to do sports activity or exercise	0	1	2	3	4
4. It is hard for me to lift something heavy	0	1	2	3	4
5. It is hard for me to take a bath or shower by myself	0	1	2	3	4
6. It is hard for me to do chores around the house	0	1	2	3	4
7. I hurt or ache	0	1	2	3	4
8. I have low energy	0	1	2	3	4

About My Feelings *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. I feel afraid or scared	0	1	2	3	4
2. I feel sad or blue	0	1	2	3	4
3. I feel angry	0	1	2	3	4
4. I have trouble sleeping	0	1	2	3	4
5. I worry about what will happen to me	0	1	2	3	4

How I Get Along with Others *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. I have trouble getting along with other kids	0	1	2	3	4
2. Other kids do not want to be my friend	0	1	2	3	4
3. Other kids tease me	0	1	2	3	4
4. I cannot do things that other kids my age can do	0	1	2	3	4
5. It is hard to keep up when I play with other kids	0	1	2	3	4

About School *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard to pay attention in class	0	1	2	3	4
2. I forget things	0	1	2	3	4
3. I have trouble keeping up with my schoolwork	0	1	2	3	4
4. I miss school because of not feeling well	0	1	2	3	4
5. I miss school to go to the doctor or hospital	0	1	2	3	4

ID# _____

Date:_____

PedsQL™

Pediatric Quality of Life Inventory

Version 4.0

PARENT REPORT for **CHILDREN** (ages **8-12**)

DIRECTIONS

On the following page is a list of things that might be a problem for **your child**. Please tell us **how much of a problem** each one has been for **your child** during the **past ONE month** by circling:

0 if it is **never** a problem
1 if it is **almost never** a problem
2 if it is **sometimes** a problem
3 if it is **often** a problem
4 if it is **almost always** a problem

There are no right or wrong answers.
If you do not understand a question, please ask for help.

*In the past **ONE month,** how much of a **problem** has your child had with …*

PHYSICAL FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Walking more than one block	0	1	2	3	4
2. Running	0	1	2	3	4
3. Participating in sports activity or exercise	0	1	2	3	4
4. Lifting something heavy	0	1	2	3	4
5. Taking a bath or shower by him or herself	0	1	2	3	4
6. Doing chores around the house	0	1	2	3	4
7. Having hurts or aches	0	1	2	3	4
8. Low energy level	0	1	2	3	4

EMOTIONAL FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Feeling afraid or scared	0	1	2	3	4
2. Feeling sad or blue	0	1	2	3	4
3. Feeling angry	0	1	2	3	4
4. Trouble sleeping	0	1	2	3	4
5. Worrying about what will happen to him or her	0	1	2	3	4

SOCIAL FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Getting along with other children	0	1	2	3	4
2. Other kids not wanting to be his or her friend	0	1	2	3	4
3. Getting teased by other children	0	1	2	3	4
4. Not able to do things that other children his or her age can do	0	1	2	3	4
5. Keeping up when playing with other children	0	1	2	3	4

SCHOOL FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Paying attention in class	0	1	2	3	4
2. Forgetting things	0	1	2	3	4
3. Keeping up with schoolwork	0	1	2	3	4
4. Missing school because of not feeling well	0	1	2	3	4
5. Missing school to go to the doctor or hospital	0	1	2	3	4

ID# _____

Date:_____

PedsQL™

Pediatric Quality of Life Inventory

Version 4.0

YOUNG CHILD REPORT (ages **5-7**)

Instructions for interviewer:

I am going to ask you some questions about things that might be a problem for some children. I want to know how much of a problem any of these things might be for you.

Show the child the template and point to the responses as you read.

If it is <u>not at all</u> a problem for you, point to the smiling face

If it is <u>sometimes</u> a problem for you, point to the middle face

If it is a problem for you <u>a lot</u>, point to the frowning face

I will read each question. Point to the pictures to show me how much of a problem it is for you. Let's try a practice one first.

	Not at all	Sometimes	A lot
Is it hard for you to snap your fingers	☺	😐	☹

Ask the child to demonstrate snapping his or her fingers to determine whether or not the question was answered correctly. Repeat the question if the child demonstrates a response that is different from his or her action.

Think about how you have been doing for the last few weeks. Please listen carefully to each sentence and tell me how much of a problem this is for you.

After reading the item, gesture to the template. If the child hesitates or does not seem to understand how to answer, read the response options while pointing at the faces.

PHYSICAL FUNCTIONING ***(problems with…)***	**Not at all**	**Some-times**	**A lot**
1. Is it hard for you to walk	0	2	4
2. Is it hard for you to run	0	2	4
3. Is it hard for you to play sports or exercise	0	2	4
4. Is it hard for you to pick up big things	0	2	4
5. Is it hard for you to take a bath or shower	0	2	4
6. Is it hard for you to do chores (like pick up your toys)	0	2	4
7. Do you have hurts or aches (***Where?_____***)	0	2	4
8. Do you ever feel too tired to play	0	2	4

Remember, tell me how much of a problem this has been for you for the last few weeks.

EMOTIONAL FUNCTIONING ***(problems with…)***	**Not at all**	**Some-times**	**A lot**
1. Do you feel scared	0	2	4
2. Do you feel sad	0	2	4
3. Do you feel mad	0	2	4
4. Do you have trouble sleeping	0	2	4
5. Do you worry about what will happen to you	0	2	4

SOCIAL FUNCTIONING ***(problems with…)***	**Not at all**	**Some-times**	**A lot**
1. Is it hard for you to get along with other kids	0	2	4
2. Do other kids say they do not want to play with you	0	2	4
3. Do other kids tease you	0	2	4
4. Can other kids do things that you cannot do	0	2	4
5. Is it hard for you to keep up when you play with other kids	0	2	4

SCHOOL FUNCTIONING ***(problems with…)***	**Not at all**	**Some-times**	**A lot**
1. Is it hard for you to pay attention in school	0	2	4
2. Do you forget things	0	2	4
3. Is it hard to keep up with schoolwork	0	2	4
4. Do you miss school because of not feeling good	0	2	4
5. Do you miss school because you have to go to the doctor's or hospital	0	2	4

How much of a problem is this for you?

Not at all	Sometimes	A lot
		

ID# _____

Date: _____

PedsQL™

Pediatric Quality of Life Inventory

Version 4.0

PARENT REPORT for **YOUNG CHILDREN** (ages 5-7)

DIRECTIONS

On the following page is a list of things that might be a problem for **your child**. Please tell us **how much of a problem** each one has been for **your child** during the **past ONE month** by circling:

0 if it is **never** a problem
1 if it is **almost never** a problem
2 if it is **sometimes** a problem
3 if it is **often** a problem
4 if it is **almost always** a problem

There are no right or wrong answers.
If you do not understand a question, please ask for help.

*In the past **ONE month**, how much of a **problem** has your child had with …*

PHYSICAL FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Walking more than one block	0	1	2	3	4
2. Running	0	1	2	3	4
3. Participating in sports activity or exercise	0	1	2	3	4
4. Lifting something heavy	0	1	2	3	4
5. Taking a bath or shower by him or herself	0	1	2	3	4
6. Doing chores, like picking up his or her toys	0	1	2	3	4
7. Having hurts or aches	0	1	2	3	4
8. Low energy level	0	1	2	3	4

EMOTIONAL FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Feeling afraid or scared	0	1	2	3	4
2. Feeling sad or blue	0	1	2	3	4
3. Feeling angry	0	1	2	3	4
4. Trouble sleeping	0	1	2	3	4
5. Worrying about what will happen to him or her	0	1	2	3	4

SOCIAL FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Getting along with other children	0	1	2	3	4
2. Other kids not wanting to be his or her friend	0	1	2	3	4
3. Getting teased by other children	0	1	2	3	4
4. Not able to do things that other children his or her age can do	0	1	2	3	4
5. Keeping up when playing with other children	0	1	2	3	4

SCHOOL FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Paying attention in class	0	1	2	3	4
2. Forgetting things	0	1	2	3	4
3. Keeping up with school activities	0	1	2	3	4
4. Missing school because of not feeling well	0	1	2	3	4
5. Missing school to go to the doctor or hospital	0	1	2	3	4

PedsQL™ Administration Guidelines℠

The following guidelines are intended for use by individuals trained in the administration of standardized questionnaires. The PedsQL™ administrator is crucial in developing rapport with the respondents, emphasizing the importance of the questionnaire, addressing concerns, and ensuring that the PedsQL™ is completed accurately and confidentially.

General Protocol

1. Create a procedure for assigning identification numbers that will allow for parent/child comparisons as well as comparisons of baseline/follow-up data.
2. If feasible, the PedsQL™ should be completed *before* the respondents complete any other health data forms and *before* they see their physician or healthcare provider.
3. The parent/child should first complete the PedsQL™ Generic Core Scales and then complete any additional PedsQL™ Module.
4. Parents, Children (8-12) and Teens (13-18) may self-administer the PedsQL™ after introductory instructions from the administrator. If the administrator determines that the child or teen is unable to self-administer the PedsQL™ (e.g., due to illness, fatigue, reading difficulties), the PedsQL™ should be read aloud to the child or teen. For the Young Child (5-7), the PedsQL™ should be administered by reading the instructions and each item to the young child word for word. At the beginning of each subscale repeat the recall interval instructions (one month or 7 days) to remind the young child to respond only for that specific recall interval. Use the separate page with the three faces response choices to help the young child understand how to answer. When reading items aloud to a child, intonation should be kept neutral to avoid suggesting an answer.
5. If a child has difficulty understanding the age-appropriate PedsQL™, the preceding age group version may be administered to the child (e.g., administering the Young Child (5-7) Self-Report version with the three faces response choices to an 8 year old). However, if a child presents with severe cognitive impairments (as determined by the administrator), the PedsQL™ may not be appropriate for that child. In such cases, only the Parent-Proxy Report should be administered to the child's parent.
6. The parent and child must complete the questionnaires *independently* of one another. Discourage the parent, child, or other family members from consulting with one another during the completion of the questionnaire. Let them know that they can feel free to discuss their answers following completion of the questionnaires, but that it is important to get both the parent's and the child's *individual* perspectives. If you are administering the questionnaire to the child, the child should be facing away from the parent.
7. If the child or parent has a question about what an item means or how they should answer it, do not interpret the question for them. Repeat the item to them verbatim. Ask them to answer the item according to what *they think the question means*. If they have trouble deciding on an answer, ask them to choose the response that comes closest to how they feel. The child and/or the parent has the option of not answering a question if they truly do not understand the question.
8. If a parent/child asks you to interpret the responses, tell her/him that you are not trained to interpret or provide a score for the answers given. If the PedsQL™ is being used for a clinical study, let the parent/child know that their answers will be combined with other participants' answers and analyzed as a group rather than as individual respondents.
9. Document all reasons for refusals and non-completions of the PedsQL™.

Administering the PedsQL™

1. The following scripts have been developed as a guide to introduce the PedsQL™ to the child and his/her parent(s). Modify the language to a style that is most appropriate for you and the respondent.

   For the child:

   The PedsQL™ asks you questions about how you feel and what you think about your health. It is not a test, and there are no right or wrong answers. It takes about 5 minutes to complete. If you have any questions, please let me know.

   For the parent:

   The PedsQL™ is a questionnaire that assesses health-related quality of life in children and adolescents. It contains questions about your child's physical, emotional, social, and school functioning ***in the past one month*** *(or for the Acute version,* ***in the past 7 days)****.*

   The PedsQL™ is brief and typically takes less than 5 minutes to complete. It is not a test, and there are no right or wrong answers. Please be sure to read the instructions carefully and choose the response that is the closest to how you truly feel. Please do not compare your answers with your child's responses. We are interested in your and your child's ***individual*** *perspectives. However, feel free to discuss the questionnaire with your child* ***after*** *you have both completed it and returned it to me. If you have any questions, please let me know.*

2. Provide the respondent with a pen or pencil and a solid writing surface. If a table is not available, the participant should be provided with an item such as a clipboard. Remain nearby should questions or concerns arise.
3. When the parent/child returns the PedsQL™, look it over and check to see that all answers have been completed. Verify that no item has more than one response. If any responses are incomplete, illegible, or there are multiple responses for an item, please ask the parent or child to indicate their response.
4. Ask the participants if they had any difficulties completing the questionnaire or if they have any other comments regarding the questionnaire. Document any important feedback.
5. Thank the parent and child for taking the time to complete the questionnaire. If the study design involves following up with these respondents, let them know that they may be asked to complete the PedsQL™ again at another time. Indicate when they can expect to be contacted again if known.

Appendix 20: Study Protocol

Standardised Testing Protocol

Preparation for Testing

1. Participant wearing comfortable clothing, ideally shorts or loose trousers, tee shirt under tight tops
2. Shoes suitable for activity
3. Patients should not have exercised vigorously within 2 hours of beginning the test
4. Encourage child to bring in their own water, or have a small drink of water ready
5. Equipment set up
6. Covid check as per CHI guidelines at Crumlin/at Blanch/at Temple Street
7. Parent/Child any questions?
8. Aim total testing time: Physical Testing + Questionnaires = 90 minutes

Standardised Testing Protocol

Parent and Child Questionnaires

1. PedsQL™ (Parent and age specific Child) Core
2. PedsQL™ (Parent and age specific Child) Pain
3. PedsQL™ (Parent and age specific Child) Multidimensional Fatigue
4. Physical Activity (child only) PAQ-C or A
    i) Child (PAQ-C 1st class to 6th class National School)
    ii)Adolescent (PAQ-A Secondary level)

Setting Up

- Child at table
- Parent in chair with clipboard
- Child facing away from Parent
- Provide with pen/pencil

PedsQL™ generic core

Setting up

- Set up table and chair for the child/teen
- Clipboard for the parent
- Child should face away from the parent
- Provide the respondents with a pen or pencil
- Remain nearby should questions or concerns arise

Administrator Role

Administrator role is to develop rapport with the respondents, emphasis of the importance of the questionnaire, address concerns and ensure that the questionnaires are completed accurately and confidentially.

2.2.3 Procedure:

1. Create a procedure for assigning identification numbers that will allow for parent/child comparisons as well as comparisons of baseline/follow-up data.

2. If feasible, the PedsQL™ should be completed before the respondents complete any other health data forms and before they see their physician or healthcare provider. The parent/child should first complete the PedsQL™ Generic Core Scales and then complete any additional PedsQL™ Module.

3. Parents, Children (8-12) and Teens (13-18) may self-administer the PedsQL™ after introductory instructions from the administrator.

4. If the administrator determines that the child or teen is unable to self-administer the PedsQL (e.g., due to illness, fatigue, reading difficulties), the PedsQL should be read aloud to the child or teen.

5. For the Young Child (5-7), the PedsQL™ should be administered by reading the instructions and each item to the young child word for word.

6. At the beginning of each subscale repeat the recall interval instructions (one month) to remind the young child to respond only for that specific recall interval.

7. Use the separate page with the three faces response choices to help the young child understand how to answer.

8. When reading items aloud to a child, intonation should be kept neutral to avoid suggesting an answer.

9. If a child has difficulty understanding the age appropriate PedsQL, the preceding age group version may be administered to the child (e.g., administering the Young Child (5-7) Self-Report version with the three faces response choices to an 8-year-old).

10. However, if a child presents with severe cognitive impairments (as determined by the administrator), the PedsQL may not be appropriate for that child. In such cases, only the Parent-Proxy Report should be administered to the child's parent.

11. . The parent and child must complete the questionnaires independently of one another.

12. Discourage the parent, child, or other family members from consulting with one another during the completion of the questionnaire.

13. Let them know that they can feel free to discuss their answers following completion of the questionnaires, but that it is important to get both the parent's and the child's individual perspectives.

14. If you are administering the questionnaire to the child, the child should be facing away from the parent.

15. If the child or parent has a question about what an item means or how they should answer it, do not interpret the question for them.

16. Repeat the item to them verbatim.

17. Ask them to answer the item according to what they think the question means.

18. If they have trouble deciding on an answer, ask them to choose the response that comes closest to how they feel.

19. The child and/or the parent has the option of not answering a question if they truly do not understand the question.

20. If a parent/child asks you to interpret the responses, tell her/him that you are not trained to interpret or provide a score for the answers given.

21. If the PedsQL™ is being used for a clinical study, let the parent/child know that their answers will be combined with other participants' answers and analysed as a group rather than as individual respondents.

22. Document all reasons for refusals and non-completions of the PedsQL™.

Administering the PedsQL™

The following scripts have been developed as a guide to introduce the PedsQL™ to the child and his/her parent(s).
Modify the language to a style that is most appropriate for you and the respondent.

For the child:

The PedsQL™ asks you questions about how you feel and what you think about your health.
It is not a test, and there are no right or wrong answers.
It takes about 5 minutes to complete.
If you have any questions, please let me know.

For the parent:

The PedsQL™ is a questionnaire that assesses health-related quality of life in children and adolescents.
It contains questions about your child's physical, emotional, social, and school functioning in the past one month (or for the Acute version, in the past 7 days).
The PedsQL™ is brief and typically takes less than 5 minutes to complete.
It is not a test, and there are no right or wrong answers.
Please be sure to read the instructions carefully and choose the response that is the closest to how you truly feel.
Please do not compare your answers with your child's responses.
We are interested in your and your child's individual perspectives.
However, feel free to discuss the questionnaire with your child after you have both completed it and returned it to me.
If you have any questions, please let me know.

When respondents have finished:

- When the parent/child returns the PedsQL™, look it over and check to see that all answers have been completed

- Verify that no item has more than one response

- If any responses are incomplete, illegible, or there are multiple responses for an item, please ask the parent or child to indicate their response

Standardised Testing Protocol

- Ask the participants if they had any difficulties completing the questionnaire or if they have any other comments regarding the questionnaire. Document any important feedback

- Thank the parent and child for taking the time to complete the questionnaire.

- If the study design involves following up, indicate when they can expect to be contacted again if known.

PedsQL™ Paediatric Pain

Equipment and Administrator Role

- Standard box of 8 crayons
- Set up table and chair for the child/teen
- Clipboard for the parent
- Child should face away from the parent
- Provide the respondents with a pen or pencil
- Remain nearby should questions or concerns arise

Procedure:

1. Self-rated questionnaire or interview format

2. VAS is a 10-cm horizontal line, anchored with descriptors "not hurting" or "no pain" and "hurting a whole lot" or "severe pain" and person marks a vertical line on the VAS for present pain and the VAS for worst pain for the previous week

3. Body outline is gender-neutral front and back whole-body drawing and 4 boxes underneath descriptive categories of pain intensity ("none, mild, moderate, severe")

4. Given a standard set of 8 colours, person selects colours to match pain intensities (colouring the 4 boxes with selected colours) and then colours in the body outline with the selected colour/intensity match

5. Children younger than 7 years will usually need to be read instructions for completing the VAS and body outline.

PedsQL™ Multidimensional Fatigue Scale

Equipment and Administration Role

- Set up table and chair for the child/teen
- Clipboard for the parent
- Child should face away from the parent
- Provide the respondents with a pen or pencil
- Remain nearby should questions or concerns arise

Standardised Testing Protocol

Procedure:

1. The 18-item PedsQL™ Multidimensional Fatigue Scale encompasses
2. General Fatigue (6 items, e.g., "I feel tired."; "I feel too tired to do things that I like to do.")
3. Sleep/Rest Fatigue (6 items, e.g., "I feel tired when I wake up in the morning."; "I rest a lot.")
4. Cognitive Fatigue (6 items, e.g., "It is hard for me to keep my attention on things."; "It is hard for me to remember what people tell me.")
5. The format, instructions, Likert scale, and scoring method are identical to the PedsQL™ 4.0 Generic Core Scales, with higher scores indicating better HRQOL (fewer problems or symptoms)

Physical Activity Questionnaire- Child (PAQ-C)

Equipment and Administrator Role

- Laminated copy for the younger children (you may need to read along with the child)

Procedure:

1. Explain it is NOT A TEST
2. Explain you are interested in ACTUAL activity during the last 7 DAYS
3. **When respondents have finished:**

- To prevent missing Data quickly glance through the questionnaires when it is gathered
- Missing one response for an activity on item 1 has little effect on the overall score, but you don't want the child missing entire items (i.e. not having a response for item 6).
- Explain to the child that you are not looking at their activity levels, but rather just making sure they haven't missed any of the questions.

Physical Activity Questionnaire – Adolescents (PAQ-A)

Procedure:

1. Explain it is NOT A TEST
2. Explain you are interested in ACTUAL activity during the last 7 DAYS
3. **When respondents have finished:**

- To Prevent Missing Data quickly glance through the questionnaires when they are gathered from the participant
- Missing one response for an activity on item 1 has little effect on the overall score, but you don't want the students missing entire items (i.e., not having a response for item 6).
- Explain to the participant that you are not looking at their activity levels, but rather just making sure they haven't missed any of the questions.

Standardised Testing Protocol

Objective Testing

Height
Procedure:

1. The participant was instructed to stand with their feet flat on the floor, touching the base of the vertical measuring column, the participant's arms were relaxed, with their bottom and shoulders touching the stadiometer.
2. The Principal Investigator lowered the horizontal bar to the head crown with hair compressed and took the measurement to the nearest 0.1 cm.

Weight
Procedure:

1. The participant was instructed to step onto the center of the weighing scales with the body weight evenly distributed between both feet until the measurement remained stable.
2. Measurement taken to the nearest 0.1kg.

Leg Length
Procedure:

1. Leg length was measured by placing a mark with a fine tipped marker on the participant's inferior aspect of each anterior superior iliac spine (ASIS) and on the most distal portion of each lateral malleolus.
2. The participant was instructed to lift their hips off the table, the legs were passively straightened to equalize the pelvis (194).
3. The participant was asked to place their hands upon the mark on the ASIS (figure N). The participant's right and left limb length was measured from the anterior superior iliac spine to the most distal portion of the lateral malleolus with a tape measure (194).
4. The measurements were taken to the nearest 0.1cm.

Functional Capacity
Six-Minute Walk Test

Procedure:

Standardised Testing Protocol

- Assemble all necessary equipment.
- The six-minute walk test (6MWT) will be conducted according to the American Thoracic Society (ATS) guidelines
- The primary and only outcome measured in this 6MWT is distance walked
- Pulse rate and oxygen saturations are usually recorded for patients with respiratory disease, but this will be unnecessary for patients included in this study and will not be assessed
- Perceived level of breathlessness will be recorded as per the protocol
- A "warm-up" period before the test should not be performed
- The patient should sit at rest in a chair, located near the starting position, for at least 10 minutes before the test starts
- During this time and make sure that clothing and shoes are appropriate
- Set the lap counter to zero and the timer to 6 minutes

Instruct the patient as follows

Borg Scale

At the beginning of the 6-minute exercise, show the scale to the patient and ask the patient this:

"Please (grade) your level of shortness of breath using this scale."

Then ask this: "Please grade your level of fatigue using this scale."

"The object of this test is to walk as far as possible for 6 minutes. You will walk back and forth in this hallway. You are permitted to slow down, to stop, and to rest as necessary. You may lean against the wall while resting but resume walking as soon as you are able. You will be walking back and forth around the cones. You should pivot briskly around the cones and continue back the other way without hesitation. Now I'm going to show you. Please watch the way I turn without hesitation.

- Demonstrate by walking one lap yourself. Walk and pivot around a cone briskly.

"Are you ready to do that? I am going to use this counter to keep track of the number of laps you complete. I will click it each time you turn around at this starting line. Remember that the object is to walk AS FAR AS POSSIBLE for 6 minutes, but don't run or jog. Start now, or whenever you are ready."

- Position the patient at the starting line.
- You should also stand near the starting line during the test.
- Do not walk with the patient.
- As soon as the patient starts to walk, start the timer.
- Do not talk to anyone during the walk.
- Use an even tone of voice when using the standard phrases of encouragement.
- Watch the patient.
- Do not get distracted and lose count of the laps.

Standardised Testing Protocol

- Each time the participant returns to the starting line, click the lap counter once (or mark the lap on the worksheet).
- Let the participant see you do it.
- Exaggerate the click using body language, like using a stopwatch at a race.
- After the first minute, tell the patient the following (in even tones)

"You are doing well. You have 5 minutes to go." When the timer shows 4 minutes remaining, tell the patient the following: "Keep up the good work. You have 4 minutes to go." When the timer shows 3 minutes remaining, tell the patient the following: "You are doing well. You are halfway done." When the timer shows 2 minutes remaining, tell the patient the following: "Keep up the good work. You have only 2 minutes left." When the timer shows only 1-minute remaining, tell the patient: "You are doing well. You have only 1 minute to go."

- Do not use other words of encouragement (or body language to speed up)
- If the patient stops walking during the test and needs a rest, say this:

"You can lean against the wall if you would like; then continue walking whenever you feel able."

- Do not stop the timer.
- If the patient stops before the 6 minutes are up and refuses to continue (or you decide that they should not continue), wheel the chair over for the patient to sit on, discontinue the walk, and note on the worksheet the distance, the time stopped, and the reason for stopping prematurely.
- When the timer is 15 seconds from completion, say this:

"In a moment I'm going to tell you to stop. When I do, just stop right where you are, and I will come to you."

When the timer rings (or buzzes), say this: "Stop!" Walk over to the patient.

- Consider taking the chair if they look exhausted

- Mark the spot where they stopped by placing a bean bag or a piece of tape on the floor
- Record the post walk Borg dyspnea and fatigue levels and ask this:

"What, if anything, kept you from walking farther?

- Record the number of laps from the counter (or tick marks on the worksheet)

- Record the additional distance covered (the number of meters in the final partial lap) using the markers on the wall as distance guides

- Calculate the total distance walked, rounding to the nearest meter, and record it on the worksheet; test results were normalized for leg length
- Congratulate the patient on good effort and offer a drink of water.

Standardised Testing Protocol

Dynamic Balance
Y Balance Test

Procedure:

1. The participants conducted the YBT following the six-minute walk test, which acted as a warmup.
2. The participant was instructed to remove their shoes and socks.
3. The YBT short video (51 seconds) was shown to the participant, and they were given the opportunity to ask questions (Appendix N). They were informed they could stop at any time, and to alert the Principal Investigator if they experienced any pain or discomfort or dizziness, nausea, or malaise during the test. This was explained using age-appropriate language.
4. Verbal, standardised instructions were provided to the participant. Language was adapted for younger participants as appropriate. The participant was directed to stand on the platform with their toes behind the line and to push the reach box in the red target area, in the direction that we were testing. Stance foot movement and body movement was also allowed under control otherwise it was too difficult to standardise the amount of movement allowed.
5. The test started with six practice trials in each leg in each direction following the standard testing order, alternating each leg. The practice trial distance was not recorded. The trail was discarded and repeated if the participant failed to maintain unilateral stance or failed to maintain contact with the reach box (e.g., kicked or flicked the reach box), or used the reach box for support (e.g., placed foot on top of the reach box).
6. Following the practice trials, the test was conducted. The reach direction was as follows: i) Right Anterior, ii) Left Anterior, iii) Right Posteromedial iv) Left Posteromedial v) Right posterolateral and vi) Left posterolateral.
7. Three trials in each reach direction were recorded to the nearest 0.5cm. The maximum reach distance in each direction was recorded.

Standardised Testing Protocol

Hypermobility Screening Assessments

Lower Limb Assessment Scale

Procedure:

- Use Lower Limb Assessment Scale score sheet.
- Providing a score to a maximum of 12 marks for each limb
- To score**, each limb** is calculated separately giving a left score and right score.
- Each YES is given one mark
- A total of score of 12 marks is available.

Beighton Score

Procedure:

Description	Bilateral Testing	Score
Passive dorsiflexion of the fifth metacarpophalangeal joint to ≥ 90degrees	YES	
Passive hyperextension of the elbow ≥10 degrees	YES	
Passive hyperextension of the knee ≥10 degrees	YES	
Passive apposition of the thumb to the flexor side of the forearm, while shoulder is flexed to 90 degrees, elbow extended, and hand pronated	YES	
Forward flexion of the trunk, with knees straight hand palms rest easily on floor	NO	
Total score /9		

One point may be gained for each side for item 1-4 (maximum 2 per item if left and right are positive) and one point for item 5. The maximum hypermobility score is nine points.

Item 1. Passive dorsiflexion of the fifth metacarpophalangeal joint. Score is positive is ≥90 degrees (Bilateral testing)

Test position	**Motion Tested**	**Positioning Goniometer**	**Anatomical Landmarks**
Sit on a chair at the short side Of the table with arm in 80◦ abduction Elbow flexed 90 deg, forearm resting On table, forearm pronated	Passive Dorsiflexion	MCP 5	Dorsal side metacarpal 5 in length of digit 5

Standardised Testing Protocol

Item 2.
Passive hyperextension of the elbow. Score is positive if ≥10 degrees (Bilateral Testing)

Test position Landmarks	Motion Tested	Positioning Goniometer	Anatomical
Sit on a chair Shoulder 90 deg Flexion, forearm Supinated	Passive hypertext. of elbow	Lateral epicondyle humerus	Along humerus towards greater tuberosity Along radius towards radial styloid

Item 3.
Passive hyperextension of the knee. Score is positive if ≥10 degrees (Bilateral Testing)

Test position	Motion Tested	Positioning Goniometer	Anatomical Landmarks
Lying backwards With legs in Horizontal position	passive hyperextension knee	Lateral femoral epicondyle	Along femur pointed at greater trochanter. Along Fibular pointed at lateral malleolus

Item 4.
Passive apposition of the thumb to the flexor side of the forearm, while the shoulder is 90 degrees flexed, elbow extended and hand pronated. Score is positive is the whole thumb touches the flexor side of the forearm. (Bilateral testing)

Item 5.
Forward Flexion of the trunk, with knees straight. Score is positive is the hand palms rest easily on the floor.

Muscle Endurance

Timed Straight Leg Raise

Procedure:

1. The participant remained supine on the plinth and the test was explained and the participant was informed that they could stop the test at any time if they reported any pain or discomfort during the testing.
2. The Principal Investigator instructed the participant to keep their knee straight and lift their straight leg, the principal investigator demonstrated how high with their hand.
3. The participant was instructed to hold it there for as long as they could, the maximum score was 120 seconds. If the participant could maintain the position for the maximum time, the test was stopped. This was repeated for each leg.

Muscle Length

Hamstring Length

Procedure:

1. The participant remains supine with both lower limbs extended.
2. The principal investigator flexed the hip of the tested leg to 90 degrees of flexion measured by universal goniometer and maintained this angle whilst the contralateral leg stayed flat on the examination table.
3. The Principal Investigator extended the knee of the tested leg, until either the principal investigator felt slight resistance, or the participant reported a strong but tolerable stretching sensation in the hamstring musculature.
4. The KEA was measured using a universal goniometer. The axis of the goniometer was placed at the lateral epicondyle of the femur. The moving arm pointing towards the lateral malleolus and the stationary pointing towards the greater trochanter, the degree of knee flexion from terminal knee extension was measured.

Foot Posture

Foot Posture Index

Procedure:

1. The participant was instructed to stand barefoot, in their relaxed position using the standard protocol for the FPI.
2. Each item of the FPI-6 is scored between −2 and +2, with the total six items referring to positions of the forefoot, midfoot and hindfoot, and the three planes of motion: (1) talar head palpation; (2) symmetry of supra and infra lateral malleolar curvature; (3) inversion/eversion of the calcaneus; (4) prominence in the region of the talonavicular joint; (5) height of the medial longitudinal arch; (6) abduction/ adduction of the forefoot.

Strength

Grip Strength

Procedure:

1. The participant was instructed to sit with feet flat on floor, with their shoulder adducted and neutrally rotated, their elbow at 90°, forearm in neutral, wrist between 0-30°. This position was demonstrated to the participant, and any corrections were made before the test started, including changing chair size to ensure feet were flat on the floor.
2. The dynamometer was set to 2nd handle position as per the ASHT standards.
3. The principal investigator supported the dynamometer lightly at the base during the test.
4. During the test the participant was instructed using standardised instructions to squeeze as hard they could.
5. The test was repeated three times, with each the dominant and non-dominant hand, with a 30 second rest break between both hands.
6. If the participant did not maintain correct positioning during testing the measurement was discarded and the trial was repeated.

Motor Skill Proficiency
Bruininks-Oseretsky Test of Motor Proficiency Brief

Procedure:

1. The Principal Investigator instructed the participant in the procedure of the test including the following: erasing was not allowed on any item using the pencil, the participant should use his/her preferred drawing hand, preferred foot, and preferred throwing hand. If the participant was unsure about their preferential hand/foot: i)preferred drawing hand - instruct the child to draw a line on a sheet of paper using a pencil, ii) preferred throwing hand: place a tennis ball on the table and instruct the child to throw it to you and iii) leg preference: place the tennis ball on the floor and instruct examinee to kick the ball.
2. The participant was reminded of proper form mid trial, using simple reminders that allowed the participant to continue with the task whilst listening. For example, during the one -legged hips, you could say 'Jump side to side'.
3. The items were conducted in the order as directed.
4. Item 12 had two options: knee push up full push-ups. Based on the participant's performance on the other items and your knowledge of their abilities, either knee or full push-ups was chosen, whichever was more appropriate
5. The principal investigator taught the task to the participants, using verbal and visual directions as necessary. Physical demonstration was also permitted until the participant understood the task.
6. Brief support was provided to encourage correct form, when necessary.
7. Once the participant understood the task, the directions according to the administration manual were followed.
8. The principal investigator recorded the participant's performance on each item.

Appendix 21: PedsQL Paediatric Pain Questionnaire (Parent and Child)

PedsQL™

Pediatric Pain Questionnaire™

Teen Form (13-18 years of age)

Name: _____

Date: _____ **Record Number:** _____

What words would you use to describe your pain or hurt?

_____

_____

_____

1. Put a mark on the line that best shows **how you feel now**. If you have no pain or hurt, you would put a mark at the end of the line by the happy face. If you have some pain or hurt, you would put a mark near the middle of the line. If you have a whole lot of pain or hurt, you would put a mark by the sad face.

Not hurting
No discomfort
No pain

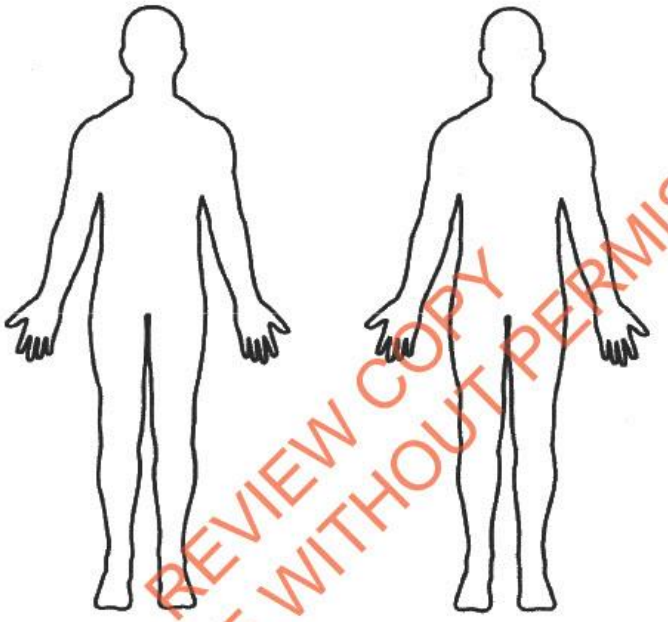
Hurting a whole lot
Very uncomfortable
Severe Pain

2. Put a mark on the line that best shows what was the **worst pain you had this week**. If you had no pain or hurt this week, you would put a mark at the end of the line by the happy face. If you had some pain or hurt, you would put a mark by the middle of the line. If the worst pain you had was a whole lot of pain, you would put a mark by the sad face.

Not hurting
No discomfort
No pain

Hurting a whole lot
Very uncomfortable
Severe Pain

Please mark an **X** on the **exact** place where you are having pain now. If there is more than one painful place, mark them '1', '2', '3', etc., starting with the most painful place as '1'.



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PedsQL PPQ-A - United States/English - MAPI Institute.
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PedsQL™

Pediatric Pain Questionnaire™

Parent of Teen Form (13-18 years of age)

Name: _____

Date: _____ **Record Number:** _____

What words would you use to describe your teen's pain or hurt?

_____

_____

_____

1. Please rate how much pain you think your teen is having **at the present time** by placing a mark somewhere on the line.

Not hurting
No discomfort
No pain

Hurting a whole lot
Very uncomfortable
Severe Pain

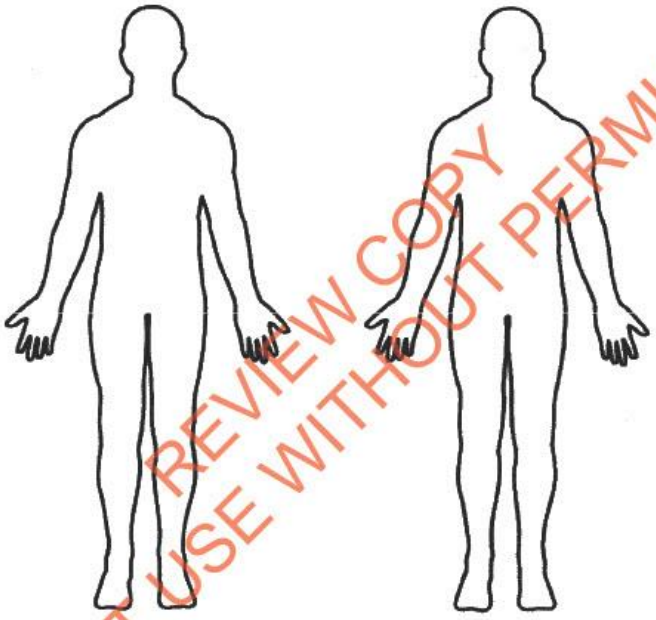
2. Please rate how severe the **worst pain** you think your teen had **in the past week** (7 days) by placing a mark somewhere on the line.

Not hurting
No discomfort
No pain

Hurting a whole lot
Very uncomfortable
Severe Pain

PedsQL PPQ-PA - United States/English - MAPI Institute.
PedsQL-3.0-PPQ-PA_AU3.0_eng-USorl.doc

Please mark an **X** on the **exact** place where you think your teen is having pain now. If there is more than one painful place, mark them '1', '2', '3', etc., starting with the most painful place as '1'.



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PedsQL PPQ-PA - United States/English - MAPI Institute.
PedsQL-3.0-PPQ-PA_AU3.0_eng-USorl.doc

PedsQL™

Pediatric Pain Questionnaire™

Child Form (**8-12** years of age)

Name: _____

Date: _____ **Record Number:** _____

What words would you use to describe your pain or hurt?

_____

_____

_____

1. Put a mark on the line that best shows **how you feel now**. If you have no pain or hurt, you would put a mark at the end of the line by the happy face. If you have some pain or hurt, you would put a mark near the middle of the line. If you have a whole lot of pain or hurt, you would put a mark by the sad face.

Not hurting
No discomfort
No pain

Hurting a whole lot
Very uncomfortable
Severe Pain

2. Put a mark on the line that best shows what was the **worst pain you had this week**. If you had no pain or hurt this week, you would put a mark at the end of the line by the happy face. If you had some pain or hurt, you would put a mark by the middle of the line. If the worst pain you had was a whole lot of pain, you would put a mark by the sad face.

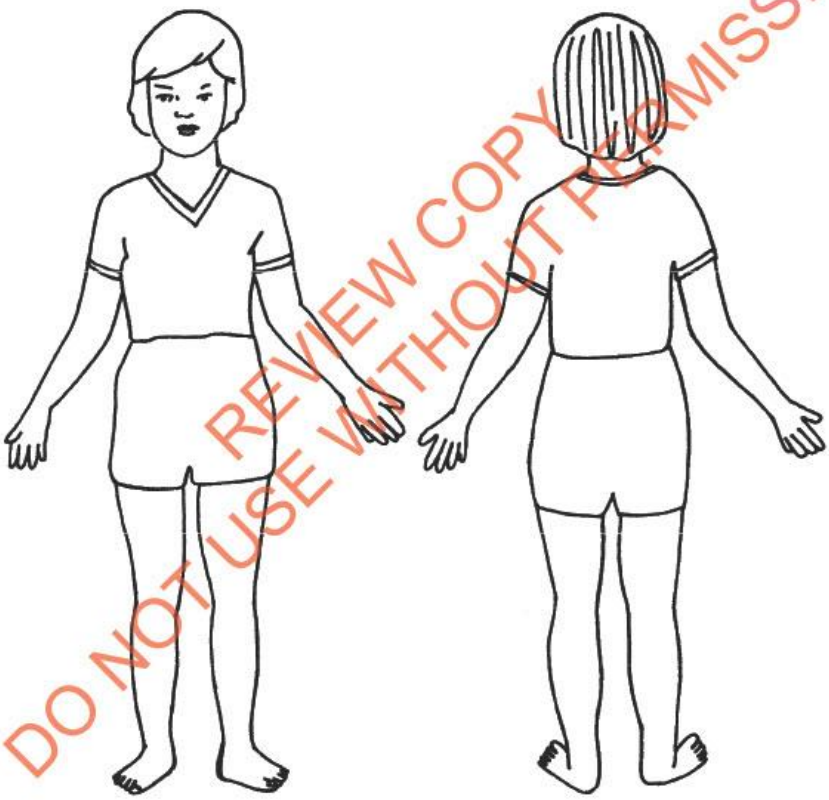
Not hurting
No discomfort
No pain

Hurting a whole lot
Very uncomfortable
Severe Pain

PedsQL PPQ-C - United States/English - MAPI Institute.
PedsQL-3.0-PPQ-C_AU3.0_eng-USorl.doc

Pick the colors that mean **No hurt, A little hurt, More hurt,** and **A lot of hurt** to you and color in the boxes. Now, using these colors, color in the body to show how you feel. Where you have no hurt, use the **No hurt** color to color in your body. If you have hurt or pain, use the color that tells how much hurt you have.

No pain No hurt	Mild pain A little hurt	Moderate pain More hurt	Severe pain A lot of hurt



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PedsQL PPQ-C - United States/English - MAPI Institute.
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PedsQL™

Pediatric Pain Questionnaire™

Parent of Child Form (**8-12** years of age)

Name: _____

Date: _____ **Record Number:** _____

What words would you use to describe your child's pain or hurt?

_____

_____

_____

1. Please rate how much pain you think your child is having **at the present time** by placing a mark somewhere on the line.

Not hurting
No discomfort
No pain

Hurting a whole lot
Very uncomfortable
Severe Pain

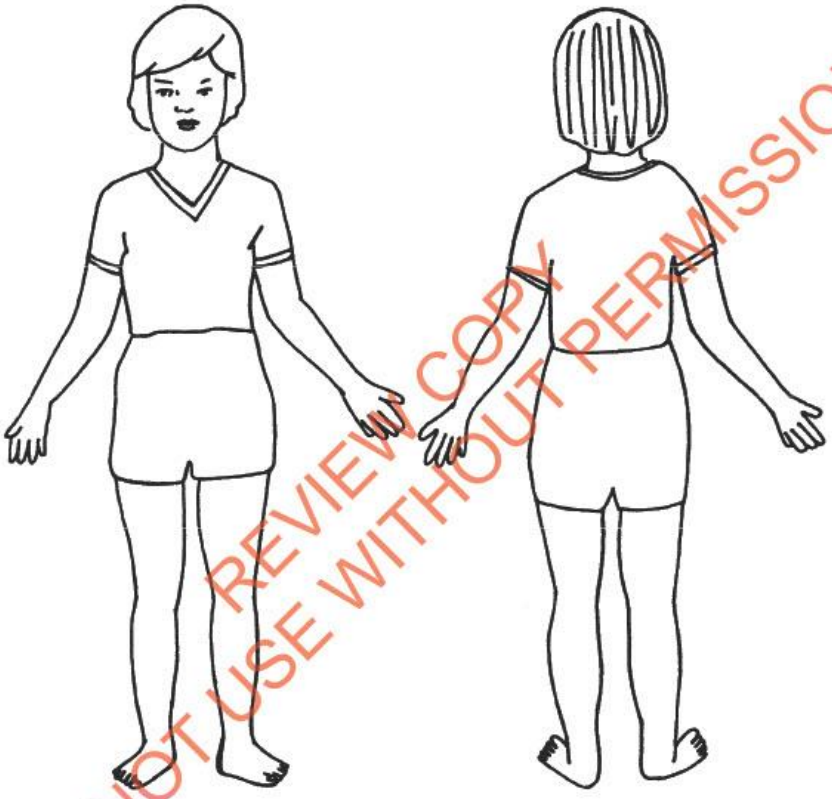
2. Please rate how severe the **worst pain** you think your child had **in the past week** (7 days) by placing a mark somewhere on the line.

Not hurting
No discomfort
No pain

Hurting a whole lot
Very uncomfortable
Severe Pain

PedsQL PPQ-PC - United States/English - MAPI Institute.
PedsQL-3.0-PPQ-PC_AU3.0_eng-USorl.doc

Please mark an **X** on the **exact** place where you think your child is having pain now. If there is more than one painful place, mark them '1', '2', '3', etc., starting with the most painful place as '1'.



PedsQL PPQ-PC - United States/English - MAPI Institute.
PedsQL-3.0-PPQ-PC_AU3.0_eng-USorl.doc

PedsQL™

Pediatric Pain Questionnaire™

Young Child Form (5-7 years of age)

Name: _____

Date: _____ **Record Number:** _____

What words would you use to describe your pain or hurt?

_____

_____

_____

1. Put a mark on the line that best shows **how you feel now**. If you have no pain or hurt, you would put a mark at the end of the line by the happy face. If you have some pain or hurt, you would put a mark near the middle of the line. If you have a whole lot of pain or hurt, you would put a mark by the sad face.

Not hurting
No discomfort
No pain

Hurting a whole lot
Very uncomfortable
Severe Pain

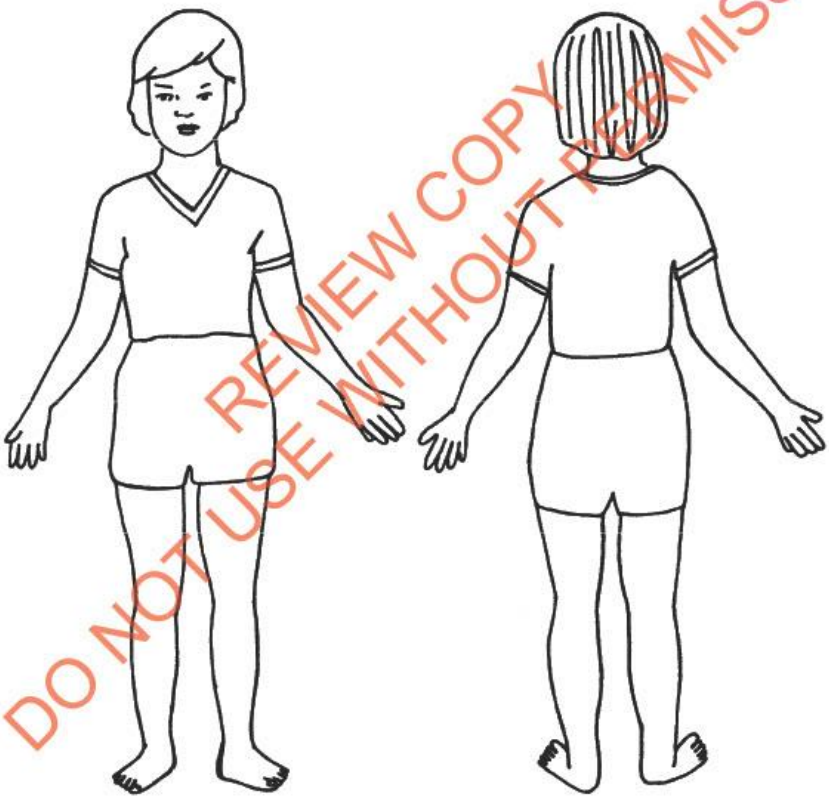
2. Put a mark on the line that best shows what was the **worst pain you had this week**. If you had no pain or hurt this week, you would put a mark at the end of the line by the happy face. If you had some pain or hurt, you would put a mark by the middle of the line. If the worst pain you had was a whole lot of pain, you would put a mark by the sad face.

Not hurting
No discomfort
No pain

Hurting a whole lot
Very uncomfortable
Severe Pain

Pick the colors that mean **No hurt, A little hurt, More hurt,** and **A lot of hurt** to you and color in the boxes. Now, using these colors, color in the body to show how you feel. Where you have no hurt, use the **No hurt** color to color in your body. If you have hurt or pain, use the color that tells how much hurt you have.

No pain No hurt	Mild pain A little hurt	Moderate pain More hurt	Severe pain A lot of hurt



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PedsQL PPQ-YC - United States/English - MAPI Institute.
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PedsQL™

Pediatric Pain Questionnaire™

Parent of Young Child Form (**5-7** years of age)

Name: _____

Date: _____ **Record Number:** _____

What words would you use to describe your child's pain or hurt?

_____

_____

_____

1. Please rate how much pain you think your child is having **at the present time** by placing a mark somewhere on the line.

Not hurting
No discomfort
No pain

Hurting a whole lot
Very uncomfortable
Severe Pain

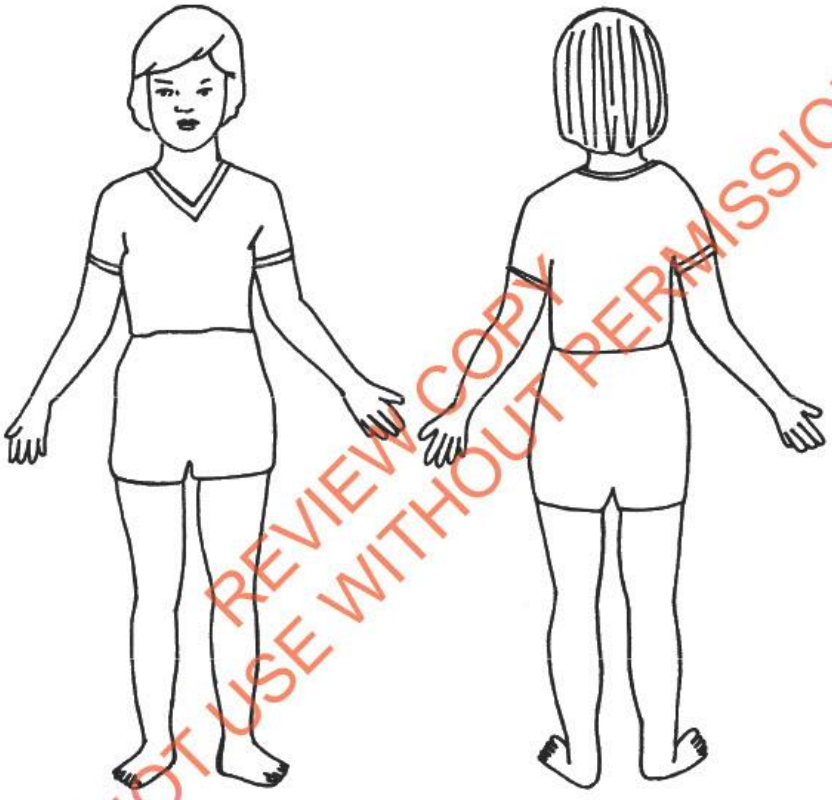
2. Please rate how severe the **worst pain** your child had **in the past week** (7 days) by placing a mark somewhere on the line.

Not hurting
No discomfort
No pain

Hurting a whole lot
Very uncomfortable
Severe Pain

PedsQL PPQ-PYC - United States/English - MAPI Institute.
PedsQL-3.0-PPQ-PYC_AU3.0_eng-USorl.doc

Please mark an **X** on the **exact** place where you think your child is having pain now. If there is more than one painful place, mark them '1', '2', '3', etc., starting with the most painful place as '1'.



Front Back

Appendix 22: PedsQL Multidimensional Fatigue Scale (Parent and Child)

ID# _____

Date: _____

PedsQL™

Multidimensional Fatigue Scale

Version 3.0 - English (United Kingdom)

TEEN REPORT (ages 13-18)

DIRECTIONS

On the following page is a list of things that might be a problem for you. Please tell us **how much of a problem** each one has been for you during the **past ONE month** by circling:

0 if it is **never** a problem
1 if it is **almost never** a problem
2 if it is **sometimes** a problem
3 if it is **often** a problem
4 if it is **almost always** a problem

There are no right or wrong answers.
If you do not understand a question, please ask for help.

*In the past **ONE month**, how much of a **problem** has this been for you …*

GENERAL FATIGUE *(problems with...)*	Never	Almost Never	Some-times	Often	Almost Always
1. I feel tired	0	1	2	3	4
2. I feel physically weak (not strong)	0	1	2	3	4
3. I feel too tired to do things that I like to do	0	1	2	3	4
4. I feel too tired to spend time with my friends	0	1	2	3	4
5. I have trouble finishing things	0	1	2	3	4
6. I have trouble starting things	0	1	2	3	4

SLEEP/REST FATIGUE *(problems with...)*	Never	Almost Never	Some-times	Often	Almost Always
1. I sleep a lot	0	1	2	3	4
2. It is hard for me to sleep through the night	0	1	2	3	4
3. I feel tired when I wake up in the morning	0	1	2	3	4
4. I rest a lot	0	1	2	3	4
5. I take a lot of naps	0	1	2	3	4
6. I spend a lot of time in bed	0	1	2	3	4

MENTAL FATIGUE *(problems with...)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard for me to keep my attention on things	0	1	2	3	4
2. It is hard for me to remember what people tell me	0	1	2	3	4
3. It is hard for me to remember what I just heard	0	1	2	3	4
4. It is hard for me to think quickly	0	1	2	3	4
5. I have trouble remembering what I was just thinking	0	1	2	3	4
6. I have trouble remembering more than one thing at a time	0	1	2	3	4

Appendix 23: Physical Activity Questionnaire for Older Children (PAQ-C) and Adolescents (PAQ-A)

The Physical Activity Questionnaire for Older Children (PAQ-C) and Adolescents (PAQ-A) Manual

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TABLE OF CONTENTS

CHAPTER 1: INTRODUCTION

"*An important challenge in determining the relationship between health and physical activity is valid assessment.*"

Various levels of physical activity participation are associated with health benefits and/or health risks. As a result, it is important that we have valid tools for assessing physical activity at various ages. This becomes particularly important with longitudinal research, which might span a number of years. The Physical Activity Questionnaire for Older Children (PAQ-C) and the Physical Activity Questionnaire for Adolescents (PAQ-A) provide a general measure of physical activity for youth from grades 4-12 (approximately ages 8-20).

The purpose of the PAQ manual is to ensure that you can easily administer the PAQ measures in research and to provide you with a library of studies utilizing the PAQ-C and the PAQ-A.

Physical Activity Questionnaire for Older Children (PAQ-C)

The PAQ-C is appropriate for elementary school-aged children (grades 4-8; approximately ages 8-14) who are currently in the school system and have recess as a regular part of their school week.

Physical Activity Questionnaire for Adolescents (PAQ-A)

The PAQ-A is appropriate for high school students (grades 9-12; approximately ages 14-20) who are currently in the school system.

This manual provides a comprehensive overview of the PAQ-C and PAQ-A.

- Chapter 1: Describes why the PAQ-C and the PAQ-A were created and the limitations and strengths of these measures.
- Chapters 2 and 3: Includes keys to successful administration of the PAQ-C and the PAQ-A, scoring the questionnaires, validation and reliability studies, and the actual measures.
- Chapter 4: Summarizes the studies that we are aware of (as of August 2004) that have used or reviewed the PAQ-C or PAQ-A.

1.1 Why were the PAQ-C and the PAQ-A Created?

It is difficult to determine the best instruments to assess physical activity when a gold standard does not exist. Examples of instruments that have been used include a variety of physiological indicators, laboratory methods, direct observation, motion sensors, and self-report measures (Sallis & Saelens, 2000; Tremblay, Shephard, McKenzie, & Gledhill, 2001; Welk & Wood, 2000). Self-report measures are most frequently utilized for the assessment of physical activity levels in children and adolescents because they are typically low in cost and can be

easily administered to large populations. However, few recall instruments have strong validity and are feasible for large-scale research (Crocker, Bailey, Faulkner, Kowalski, & McGrath, 1997).

In response to the need for a valid and feasible self-report measure for large-scale research with children and adolescents, the Physical Activity Questionnaire for Older Children (PAQ-C; Crocker, Bailey, Faulkner, Kowalski, & McGrath, 1997; Kowalski, Crocker, & Faulkner, 1997) and the Physical Activity Questionnaire for Adolescents (PAQ-A; Kowalski, Crocker, & Kowalski, 1997) were developed and validated. The PAQ-C and PAQ-A are self-administered, 7-day recall questionnaires that measure general moderate to vigorous physical activity levels during the school year. Generally, the PAQs have had relatively strong correlation coefficients with other physical activity measures compared to other recall measures (Kowalski, Crocker, & Faulkner, 1997; Kowalski, Crocker, & Kowalski, 1997).

The PAQ-C and the PAQ-A may be advantageous for use in longitudinal research. The PAQs' low cost, reliable and valid assessment of physical activity from childhood through adolescence, and ease of administration make the PAQs feasible for large-scale studies. The questionnaires use a common scoring scheme and were used successfully in the University of Saskatchewan's longitudinal bone mineral accrual study (Bailey, McKay, Mirwald, Crocker, & Faulkner, 1999).

1.2 The Limitations and Strengths of the PAQ Measures

All physical activity measures, including the PAQ-C and the PAQ-A, have their strengths and limitations. For example, some tools may or may not be feasible for large-scale research, be cost and time efficient, have good adherence, have participant demand, and/or have acceptable distribution properties.

The PAQ-C's and the PAQ-A's limitations

1) The PAQ-C and PAQ-A were developed to assess general levels of physical activity. They do not provide an estimate of caloric expenditure or specific frequency, time, and intensity information.
2) The PAQs do not discriminate between specific activity intensities, such as moderate and vigorous activities; they simply provide a summary activity score (see the scoring section in Chapters 2 and 3).
3) The PAQ-C and the PAQ-A are only appropriate when used during the school year; they should *not* be used to assess physical activity in the summer or holiday periods. Therefore, the PAQ-C and the PAQ-A only assess activities for individuals in the school system.

The PAQ-C's and the PAQ-A's strengths

1) The PAQ-C and the PAQ-A have been supported as valid and reliable measures of general physical activity levels from childhood to adolescence (see the validation/reliability studies in Chapters 2 and 3). The PAQs' measurement of general physical activity levels is one its strengths because it is difficult to precisely measure intensity, frequency, and duration of young people's activities, especially with self-report (Kowalski, Crocker, & Faulkner, 1997)
2) The PAQs utilize memory cues such as lunch and evening items to enhance the recall ability of children and adolescents (see the PAQ measures in Chapters 2 and 3).
3) The PAQ-C and PAQ-A are cost and time efficient, easy to administer to large-scale populations, and display normal distribution properties (see the validation reliability studies in Chapters 2 and 3). .

1.3 References

Bailey, D. A., McKay, H. A., Mirwald, R. L., Crocker, P. R. E., & Faulkner, R. A. (1999). A six-year longitudinal study of the relationship of physical activity to bone mineral accrual in growing children: The University of Saskatchewan bone mineral accrual study. *Journal of Bone and Mineral Research, 14*, 1672-1679.

Crocker, P. R. E., Bailey, D. A., Faulkner, R. A., Kowalski, K. C., & McGrath, R. (1997). Measuring general levels of physical activity: Preliminary evidence for the Physical Activity Questionnaire for Older Children. *Medicine and Science in Sports and Exercise, 29,* 1344-1349.

Kowalski, K. C., Crocker, P. R. E., & Faulkner, R. A. (1997). Validation of the Physical Activity Questionnaire for Older Children. *Pediatric Exercise Science, 9,* 174-186.

Kowalski, K. C., Crocker, P. R. E., & Kowalski, N. P. (1997). Convergent validity of the Physical Activity Questionnaire for Adolescents. *Pediatric Exercise Science, 9,* 342-352.

Sallis, J. F., & Saelens, B. E. (2000). Assessment of physical activity by self-report: Status, limitations, and future directions. *Research Quarterly for Exercise and Sport, 71,* 1-14.

Tremblay, M. S., Shephard, R. J., McKenzie, T. L., & Gledhill, N. (2001). Physical activity assessment options within the context of the Canadian Physical Activity, Fitness, and Lifestyle Appraisal. *Canadian Journal of Applied Physiology, 26*, 388-407.

Welk, G. J. & Wood, K. (2000). Physical activity assessments in physical education: A practical review of instruments and their use in the curriculum. *Journal of Physical Education, Recreation and Dance, 71,* 30-40.

CHAPTER 2: Physical Activity Questionnaire for Older Children (PAQ-C)

2.1 What is the PAQ-C?

The PAQ-C is a self-administered, 7-day recall instrument. *It was developed to assess general levels of physical activity throughout the elementary school year for students in grades 4 to 8 and approximately 8 to 14 years of age.* The PAQ-C can be administered in a classroom setting and provides a summary physical activity score derived from nine items, each scored on a 5-point scale.

2.2 Keys to Successful Administration

1) *When the PAQ-C is administered it is important to stress 2 points:*

a) Explain it is **NOT A TEST**

b) Explain you are interested in **ACTUAL** activity during the last 7 **DAYS**

2) *To Prevent Missing Data, have the research assistants quickly glance through the questionnaires when they are gathered from the students.*

a) Missing one response for an activity on item 1 has little effect on the overall score, but you don't want the students missing entire items (ie. not having a response for item 6).

b) Explain to the students that the research assistants are not looking at their activity levels, but rather just making sure they haven't missed any of the questions.

3) *Overhead projectors may be helpful with younger age groups.*

a) This allows researchers to read along with the students as they fill out their questionnaires.

Scoring

Overall process - Find an activity score between 1 and 5 for each item (excluding item 10)

Five Easy Steps

1) *Item 1 (Spare time activity)*

- Take the mean of all activities ("no" activity being a 1, "7 times or more" being a 5) on the activity checklist to form a composite score for item 1.

2) *Items 2 to 8 (PE, recess, lunch, right after school, evening, weekends, and describes you best)*

- The answers for each item start from the lowest activity response and progress to the highest activity response
- Simply use the reported value that is checked off for each item (the lowest activity response being a 1 and the highest activity response being a 5).

3) *Item 9*

- Take the mean of all days of the week ("none" being a 1, "very often" being a 5) to form a composite score for item 9.

4) *Item 10*

- Can be used to identify students who had unusual activity during the previous week, but this question is **NOT** used as part of the summary activity score.

5) *How to calculate the final PAQ-C activity summary score*

- Once you have a value from 1 to 5 for each of the 9 items (items 1 to 9) used in the physical activity composite score, you simply take the mean of these 9 items, which results in the final PAQ-C activity summary score.

- A score of 1 indicates low physical activity, whereas a score of 5 indicates high physical activity.

2.3 Validation Reliability Studies Concerning the PAQ-C

The following paragraphs summarize the original development, validity, and reliability studies for the PAQ-C. The summaries provide a brief synopsis of each study's findings (we recommend that the complete studies be reviewed as the final reference).

Crocker, P. R. E., Bailey, D. A., Faulkner, R. A., Kowalski, K. C., & McGrath, R. (1997). Measuring general levels of physical activity: Preliminary evidence for the Physical Activity Questionnaire for Older Children. ***Medicine and Science in Sports and Exercise, 29,*** **1344-1349.**

Evidence was provided that supported the PAQ-C as a reliable and valid measure of general physical activity levels in children during the school year. In three studies, Crocker, Bailey, Faulkner, Kowalski, and McGrath (1997) administered the PAQ-C to (N = 215, N = 84, and N = 200) elementary school children during the school year. The children were between the ages of 8 to 16 and attended a public school.

In the first study, the item and scale properties of the PAQ-C were examined. Ninety girls and 125 boys (ages 9-15) completed the PAQ-C on the same day. The mean activity score for females was 2.96 (SD = 0.69) and 3.44 (SD = 0.68) for males. Boys were significantly more active than girls with respect to the PAQ-C mean scores, t (213) = 5.15, $p < 0.01$, and each item score ($p < 0.05$), excluding the physical education item ($p < 0.08$). The item scale correlations were all above 0.30, and the scale reliability was acceptable for both females (α = 0.83) and males (α = 0.80). Recess and lunch items had the lowest correlations with the other items for males (r = 0.33 and 0.30 respectively) and females (r = 0.42 and 0.55 respectively). Most PAQ-C items had means close to the center of the range and the variability was acceptable. Overall, the PAQ-C was found to have acceptable measurement properties.

The second study examined the PAQ-C's test re-test reliability, internal consistency, and sensitivity to gender differences. Forty-three boys and 41 girls (ages 9-14) completed the PAQ-C. The children were assessed twice during school hours with one week in between assessments. The PAQ-C was relatively stable over the one-week assessment period (males, r = 0.75 and females, r = 0.82). However, further analysis showed significant increases in PAQ-C activity scores for both males, 2.85 (SD = 0.73) to 3.16 (SD = 0.91) and females, 2.56 (SD = 0.65) to 2.79 (SD = 0.80) over the two assessments, F (1,83) = 22.26, $p < 0.01$. Crocker et al. (1997) suggested a possible rationale for the increase in activity might be due to the change in weather. The first assessment week was cold and snowy, whereas the second assessment week was much warmer. The internal consistency for the first assessment was (α = 0.79) and (α = 0.89) for the second assessment. In general, the boys were found to be more active than the girls for weeks one and two, t (82) = 1.93, $p < 0.05$ and t (82) = 1.97, $p < 0.05$ respectively. The results of this study provide support for the test-retest reliability of the PAQ-C, and, similar to study 1, showed that the PAQ-C was sensitive to gender differences in physical activity levels.

The third study examined the reliability of the averages of 2 or 3 PAQ-C scores as a composite yearly activity score for children. Ninety-eight boys and 102 girls (ages 8-16) who were participants in the Saskatchewan pediatric bone study completed the PAQ-C. The PAQ-C was slightly modified for the adolescent participants with the recess item omitted, and some of

the activity checklist items were changed to represent adolescent activity choices. Using generalizability theory, the results suggested that the use of 3 and 2 PAQ-C scores as a yearly activity composite score were reliable for younger participants (G = 0.86 and G = 0.80 respectively) and older participants (G = 0.90 and G = 0.85 respectively). Sex, F (1,199) = 20.22, $p < 0.01$, and time, F (2,398) = 34.34, $p < 0.01$, effects were found. The marginal mean male activity score was higher than females' (3.11 and 2.71 respectively). Students were more active in April than Oct-Nov (3.10 and 2.79 respectively). In summary, the PAQ-C had acceptable measurement properties, internal consistency, and reliability for using the average of either two or three PAQ-C scores gathered during fall, winter, and spring. These results provided initial support that the PAQ-C is a valid measure of physical activity in children.

Kowalski, K. C., Crocker, P. R. E., & Faulkner, R. A. (1997). Validation of the Physical Activity Questionnaire for Older Children. *Pediatric Exercise Science, 9,* 174-186.

Two studies by Kowalski, Crocker, and Faulkner (1997) supported the PAQ-C as a valid measure of general physical activity levels. Two independent samples (N = 89 and N = 97) of children grades 4 to 8 completed the PAQ-C along with other physical activity measures.

In the first study, the convergent, construct, and divergent validity of the PAQ-C were examined. Thirty-eight boys and 51 girls ages 8 to 13 completed a behavioural conduct scale (M = 2.92, SD = 0.53), an athletic competence scale (M = 2.94, SD = 0.58), the PAQ-C (M = 3.23, SD = 0.78), and an activity rating (M = 3.62, SD = 1.02). Following the questionnaires, the classroom teachers completed a teacher's rating of physical activity questionnaire (M = 68.13, SD = 10.97), and the children completed the moderate to vigorous physical activity (MVPA) each day for 1 week.

Convergent validity was supported by moderate relationships with the activity rating (r = 0.63), week summation of 24-hr moderate to vigorous activity recalls (r = 0.53), and teacher's rating of physical activity (r = 0.45). The PAQ-C's moderate correlation with perceptions of athletic competence (r = 0.48) provided support for the construct validity of the PAQ-C. Divergent validity of the PAQ-C was supported by no relationship between the behavioural conduct scale and the PAQ-C. Gender differences were found on the PAQ-C and teacher's rating of physical activity.

In the second study, the convergent and construct validity of the PAQ-C was further examined. Forty-one boys and 56 girls completed the PAQ-C (M = 3.35, SD = 0.68), an activity rating (M = 3.67, SD = 0.97), the Leisure Time Exercise Questionnaire ([LTEQ]; M = 75.31, SD = 58.20), the Canadian home fitness test ([step test]; M = 4.09, SD = 1.68), the seven-day recall inteview ([PAR]; M = 37.72, SD = 4.13), and wore the Caltrac motion sensor ([Caltrac]; M = 426.54, SD = 131.61). The children completed the Caltrac and PAR during a different week from the other measures due to possible carry over effects. The PAQ-C was moderately related to the activity rating (r = 0.57), LTEQ (r = 0.41), Caltrac (r = 0.39), PAR (r = 0.46), and the step test of fitness (r = 0.28). Unlike the first study, no gender differences were found for the PAQ-C. Overall, the results of these studies supported the validity of the PAQ-C.

2.4 The PAQ-C Measure

See the following page.

Physical Activity Questionnaire (Elementary School)

Name:_____ Age:_____

Sex: M_____ F_____ Grade:_____

Teacher:_____

We are trying to find out about your level of physical activity from ***the last 7 days*** (in the last week). This includes sports or dance that make you sweat or make your legs feel tired, or games that make you breathe hard, like tag, skipping, running, climbing, and others.

Remember:

1. There are no right and wrong answers — this is not a test.
2. Please answer all the questions as honestly and accurately as you can — this is very important.

---

1. Physical activity in your spare time: Have you done any of the following activities in the past 7 days (last week)? If yes, how many times? (Mark only one circle per row.)

	No	1-2	3-4	5-6	7 times or more
Skipping	❍	❍	❍	❍	❍
Rowing/canoeing	❍	❍	❍	❍	❍
In-line skating	❍	❍	❍	❍	❍
Tag	❍	❍	❍	❍	❍
Walking for exercise	❍	❍	❍	❍	❍
Bicycling	❍	❍	❍	❍	❍
Jogging or running	❍	❍	❍	❍	❍
Aerobics	❍	❍	❍	❍	❍
Swimming	❍	❍	❍	❍	❍
Baseball, softball	❍	❍	❍	❍	❍
Dance	❍	❍	❍	❍	❍
Football	❍	❍	❍	❍	❍
Badminton	❍	❍	❍	❍	❍
Skateboarding	❍	❍	❍	❍	❍
Soccer	❍	❍	❍	❍	❍
Street hockey	❍	❍	❍	❍	❍
Volleyball	❍	❍	❍	❍	❍
Floor hockey	❍	❍	❍	❍	❍
Basketball	❍	❍	❍	❍	❍
Ice skating	❍	❍	❍	❍	❍
Cross-country skiing	❍	❍	❍	❍	❍
Ice hockey/ringette	❍	❍	❍	❍	❍
Other:					
_____	❍	❍	❍	❍	❍
_____	❍	❍	❍	❍	❍

2. In the last 7 days, during your physical education (PE) classes, how often were you very active (playing hard, running, jumping, throwing)? (Check one only.)

- I don't do PE
- Hardly ever
- Sometimes
- Quite often
- Always

3. In the last 7 days, what did you do most of the time *at recess*? (Check one only.)

- Sat down (talking, reading, doing schoolwork)
- Stood around or walked around
- Ran or played a little bit
- Ran around and played quite a bit
- Ran and played hard most of the time

4. In the last 7 days, what did you normally do *at lunch* (besides eating lunch)? (Check one only.)

- Sat down (talking, reading, doing schoolwork)
- Stood around or walked around
- Ran or played a little bit
- Ran around and played quite a bit
- Ran and played hard most of the time

5. In the last 7 days, on how many days *right after school*, did you do sports, dance, or play games in which you were very active? (Check one only.)

- None
- 1 time last week
- 2 or 3 times last week
- 4 times last week
- 5 times last week

6. In the last 7 days, on how many *evenings* did you do sports, dance, or play games in which you were very active? (Check one only.)

- None
- 1 time last week
- 2 or 3 times last week
- 4 or 5 last week
- 6 or 7 times last week

7. *On the last weekend*, how many times did you do sports, dance, or play games in which you were very active? (Check one only.)

None ..... ❍
1 time ..... ❍
2 — 3 times ..... ❍
4 — 5 times ..... ❍
6 or more times ..... ❍

8. Which *one* of the following describes you best for the last 7 days? Read *all five* statements before deciding on the *one* answer that describes you.

A. All or most of my free time was spent doing things that involve little physical effort ..... ❍

B. I sometimes (1 — 2 times last week) did physical things in my free time (e.g. played sports, went running, swimming, bike riding, did aerobics) ..... ❍

C. I often (3 — 4 times last week) did physical things in my free time ..... ❍

D. I quite often (5 — 6 times last week) did physical things in my free time ..... ❍

E. I very often (7 or more times last week) did physical things in my free time ..... ❍

9. Mark how often you did physical activity (like playing sports, games, doing dance, or any other physical activity) for each day last week.

	None	Little bit	Medium	Often	Very often
Monday	❍	❍	❍	❍	❍
Tuesday	❍	❍	❍	❍	❍
Wednesday	❍	❍	❍	❍	❍
Thursday	❍	❍	❍	❍	❍
Friday	❍	❍	❍	❍	❍
Saturday	❍	❍	❍	❍	❍
Sunday	❍	❍	❍	❍	❍

10. Were you sick last week, or did anything prevent you from doing your normal physical activities? (Check one.)

Yes ..... ❍
No ..... ❍

If Yes, what prevented you? _____

CHAPTER 3: Physical Activity Questionnaire for Adolescents (PAQ-A)

3.1 What is the PAQ-A?

The PAQ-A (a slightly modified version of the PAQ-C with the "recess" item removed) is a self-administered, 7-day recall instrument. *It was developed to assess general levels of physical activity for high school students in grades 9 to 12 and approximately 14 to 19 years of age.* The PAQ-A can be administered in a classroom setting and provides a summary physical activity score derived from eight items, each scored on a 5-point scale.

3.2 Keys to Successful Administration

1) *When the PAQ-A is administered it is important to stress 2 points:*

a) Explain it is **NOT A TEST**

b) Explain you are interested in **ACTUAL** activity during the last 7 **DAYS**

2) *To Prevent Missing Data, have the research assistants quickly glance through the questionnaires when they are gathered from the students.*

a) Missing one response for an activity on item 1 has little effect on the overall score, but you don't want the students missing entire items (ie. not having a response for item 6).

b) Explain to the students that the research assistants are not looking at their activity levels, but rather just making sure they haven't missed any of the questions.

Scoring

Overall process - Find an activity score between 1 and 5 for each item (excluding item 9)

Five Easy Steps

1) *Item 1 (Spare time activity)*

- Take the mean of all activities ("no" activity being a 1, "7 times or more" being a 5) on the activity checklist to form a composite score for item 1.

2) *Item 2 to 7 (PE, lunch, right after school, evening, weekends, describes you best)*

- The answers for each item start from the lowest activity response and progress to the highest activity response
- Simply use the reported value that is checked off for each item (the lowest activity response being a 1 and the highest activity response being a 5).

3) *Item 8*

- Take the mean of all days of the week ("none" being a 1, "very often" being a 5) to form a composite score for item 8.

4) *Item 9*

- Can be used to identify students who had unusual activity during the previous week, but this question is **NOT** used as part of the summary activity score.

5) How to calculate the final PAQ-A activity summary score

- Once you have a value from 1 to 5 for each of the 8 items (items 1 to 8) used in the physical activity composite score, you simply take the mean of these 8 items, which results in the final PAQ-A activity summary score.

- A score of 1 indicates low physical activity, whereas a score of 5 indicates high physical activity.

3.3 Validation Reliability Study Concerning the PAQ-A

The following paragraphs summarize the development, validity, and, reliability study for the PAQ-A. The summary provides a brief synopsis of the study's findings (we recommend that the complete study be reviewed as the final reference).

Kowalski, K. C., Crocker, P. R. E., & Kowalski, N. P. (1997). Convergent validity of the Physical Activity Questionnaire for Adolescents. *Pediatric Exercise Science, 9,* 342-352.

The PAQ-A (a modified version of the PAQ-C) was developed to measure general levels of physical activity in adolescents. Kowalski, Crocker, and Kowalski (1997) administered the PAQ-A along with other physical activity measures to 85 high school students during the school year. The students consisted of 41 males and 44 females (grades 8 through 12), ages 13 to 20.

Two schools were assessed separately (late March-early April and late May-early June). The assessments were scheduled over two-week periods that avoided any special school events. The students were administered the PAQ-A ($M = 2.31$, $SD = 0.63$), an activity rating ($M = 3.15$, $SD = 0.93$), Leisure Time Exercise Questionnaire ([LTEQ]; M = 54.02, SD = 30.23), Caltrac motion sensor ([Caltrac]; $M = 355.88$, $SD = 126.01$), and the 7-day physical activity recall interview ([PAR]; $M = 36.21$, $SD = 3.24$). To ensure no carry over effects, the Caltrac and PAR were administered over a different 1-week period than the other measures. The PAQ-A was the only measure sensitive to gender differences, $t(83) = 3.01$, $p < 0.05$. The males were more active than the females (mean scores of 2.52 and 2.12 respectively).

The PAQ-A was significantly correlated to all self-report measures (activity rating, $r = 0.73$; LTEQ, $r = 0.57$; and PAR, $r = 0.59$). The PAQ-A was also related to the Caltrac ($r = 0.33$). A limitation of this study was that only 56.47% of students' Caltrac data were usable. The main problem with the Caltrac devices was that the students tampered with them. The PAQ-A scores differed significantly between those who had usable Caltrac data and those that did not, $t(83) = 2.78$, $p < 0.05$. These results provided support for the convergent validity of the PAQ-A.

3.4 The PAQ-A Measure

See the following page.

Physical Activity Questionnaire (High School)

Name:_____ Age:_____

Sex: M_____ F_____ Grade:_____

Teacher:_____

We are trying to find out about your level of physical activity from ***the last 7 days*** (in the last week). This includes sports or dance that make you sweat or make your legs feel tired, or games that make you breathe hard, like tag, skipping, running, climbing, and others.

Remember:

3. There are no right and wrong answers — this is not a test.
4. Please answer all the questions as honestly and accurately as you can — this is very important.

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1. Physical activity in your spare time: Have you done any of the following activities in the past 7 days (last week)? If yes, how many times? (Mark only one circle per row.)

	No	1-2	3-4	5-6	7 times or more
Skipping	❍	❍	❍	❍	❍
Rowing/canoeing	❍	❍	❍	❍	❍
In-line skating	❍	❍	❍	❍	❍
Tag	❍	❍	❍	❍	❍
Walking for exercise	❍	❍	❍	❍	❍
Bicycling	❍	❍	❍	❍	❍
Jogging or running	❍	❍	❍	❍	❍
Aerobics	❍	❍	❍	❍	❍
Swimming	❍	❍	❍	❍	❍
Baseball, softball	❍	❍	❍	❍	❍
Dance	❍	❍	❍	❍	❍
Football	❍	❍	❍	❍	❍
Badminton	❍	❍	❍	❍	❍
Skateboarding	❍	❍	❍	❍	❍
Soccer	❍	❍	❍	❍	❍
Street hockey	❍	❍	❍	❍	❍
Volleyball	❍	❍	❍	❍	❍
Floor hockey	❍	❍	❍	❍	❍
Basketball	❍	❍	❍	❍	❍
Ice skating	❍	❍	❍	❍	❍
Cross-country skiing	❍	❍	❍	❍	❍
Ice hockey/ringette	❍	❍	❍	❍	❍
Other:					
_____	❍	❍	❍	❍	❍
_____	❍	❍	❍	❍	❍

2. In the last 7 days, during your physical education (PE) classes, how often were you very active (playing hard, running, jumping, throwing)? (Check one only.)

- I don't do PE
- Hardly ever
- Sometimes
- Quite often
- Always

3. In the last 7 days, what did you normally do *at lunch* (besides eating lunch)? (Check one only.)

- Sat down (talking, reading, doing schoolwork)
- Stood around or walked around
- Ran or played a little bit
- Ran around and played quite a bit
- Ran and played hard most of the time

4. In the last 7 days, on how many days *right after school*, did you do sports, dance, or play games in which you were very active? (Check one only.)

- None
- 1 time last week
- 2 or 3 times last week
- 4 times last week
- 5 times last week

5. In the last 7 days, on how many *evenings* did you do sports, dance, or play games in which you were very active? (Check one only.)

- None
- 1 time last week
- 2 or 3 times last week
- 4 or 5 last week
- 6 or 7 times last week

6. *On the last weekend*, how many times did you do sports, dance, or play games in which you were very active? (Check one only.)

- None
- 1 time
- 2 — 3 times
- 4 — 5 times
- 6 or more times

7. Which *one* of the following describes you best for the last 7 days? Read *all five* statements before deciding on the *one* answer that describes you.

F. All or most of my free time was spent doing things that involve little physical effort ❍

G. I sometimes (1 — 2 times last week) did physical things in my free time (e.g. played sports, went running, swimming, bike riding, did aerobics) ❍

H. I often (3 — 4 times last week) did physical things in my free time ❍

I. I quite often (5 — 6 times last week) did physical things in my free time ❍

J. I very often (7 or more times last week) did physical things in my free time ❍

8. Mark how often you did physical activity (like playing sports, games, doing dance, or any other physical activity) for each day last week.

	None	Little bit	Medium	Often	Very often
Monday	❍	❍	❍	❍	❍
Tuesday	❍	❍	❍	❍	❍
Wednesday	❍	❍	❍	❍	❍
Thursday	❍	❍	❍	❍	❍
Friday	❍	❍	❍	❍	❍
Saturday	❍	❍	❍	❍	❍
Sunday	❍	❍	❍	❍	❍

9. Were you sick last week, or did anything prevent you from doing your normal physical activities? (Check one.)

Yes ❍
No ❍

If Yes, what prevented you? _____

CHAPTER 4: Overview of Studies Using the Physical Activity Questionnaires

4.1 How has the PAQ-C and the PAQ-A been utilized in research?

The Physical Activity Questionnaire for Older Children (PAQ-C) and the Physical Activity Questionnaire for Adolescents (PAQ-A) have been used to classify children and adolescents into different activity levels (e.g., Mackelvie, McKay, Khan, & Crocker, 2001a; Kowalski, Crocker, Kowalski, 1997) and to investigate the relationship between physical activity and health outcomes (e.g., Bailey, McKay, Mirwald, Crocker, & Faulkner, 1999; Mackelvie, McKay, Khan, & Crocker, 2001b). In the following paragraphs, every study that we are aware of that has used the PAQ-C and/or the PAQ-A (as of August 2004) will be summarized (interested readers should consult the *Web of Science* citation indexes for updates). The reference for each study summary will be provided at the beginning of its summary. *The following study summaries focus on how the PAQ-C and the PAQ-A were utilized in the research.* These summaries should be considered brief overviews only, and you are encouraged to consult the primary sources when referencing these studies.

Bailey, D. A. (1997). The Saskatchewan pediatric bone mineral accrual study: Bone mineral acquisition during the growing years. *International Journal of Sports Medicine, 18,* S191-S194.

The six-year bone mineral accrual study described by Bailey (1997) examined the bone mineral accretion from childhood to adolescence. Sixty-eight elementary school boys and 72 elementary school girls were assessed. The children were recipients of Dual x-ray scans once a year and anthropomorphic measures every six months. As well, the children completed the PAQ-C and a nutrition questionnaire at least three times per year for the first three years and then twice a year every year after.

The results of Bailey's (1997) bone mineral accrual study suggest that adolescents' growth period is an important time for bone mineral accretion. The focus of this study was on physical growth; unfortunately the physical activity results were not reported. Bailey suggested that more research was needed to examine the relationship between physical activity and bone mineral accrual during adolescents' peak growth period (see Bailey, McKay, Mirwald, Crocker, & Faulkner, 1999, for the physical activity data in this study).

Bailey, D. A., McKay, H. A., Mirwald, R. L., Crocker, P. R. E., & Faulkner, R. A. (1999). A six-year longitudinal study of the relationship of physical activity to bone mineral accrual in growing children: The University of Saskatchewan bone mineral accrual study. *Journal of Bone and Mineral Research, 14*, 1672-1679.

The PAQ-C was used successfully in longitudinal research to measure children's general physical activity levels from childhood to adolescence. The above study investigated the relationship between physical activity levels and bone mineral accrual as children matured into adolescents. Two hundred and twenty-eight children were recruited from the Saskatchewan pediatric bone mineral accrual study. All children were Caucasian and 8 to 14 years of age. After 6 years of data collection, 68 boys and 72 girls remained and most provided acceptable longitudinal data for analyses. The measures administered were DEXA scans to assess bone mineral content, height and weight measurements, 24-hr recalls to assess dietary calcium, and the PAQ-C to assess general levels of physical activity (note that the PAQ-C was incorrectly called the PAC-Q in this study).

The children completed the PAQ-C at least 3 times per year for the first three years, and every additional year after the third year the PAQ-C was completed twice per year. Based on the PAQ-C mean activity score and *SD* for each age group, every child was given an age-gender specific *Z* score that signified the child's activity level as either active, average activity, or inactive. Fifteen boys and 13 girls were categorized as active, 30 boys and 27 girls were of average activity, and 15 boys and 13 girls were inactive.

Children's physical activity scores were significantly correlated with peak total body bone mineral accrual and total amount of bone mineral accumulated during the 2 years around the age of peak bone mineral content velocity ([PBMCV]; $r = 0.39$, $r = 0.40$ respectively for males; $r = 0.41$, $r = 0.38$ respectively for females; and both $p < 0.05$). For the lumbar spine, girls produced the highest correlation between physical activity and PBMCV ($r = 0.47$). The active boys and girls combined had a significantly different magnitude of total body bone mineral accrual at peak and total amount of bone mineral accumulated during the 2 years around PBMCV compared to inactive boys and girls combined.

Ball, G. D. C., Marshall, J. D., & McCargar, L. J. (2003). Fatness and fitness in obese children at low and high health risk. *Pediatric Exercise Science, 15*, 392-405.

To investigate whether or not physical activity levels predict the presence of metabolic risk factors of cardiovascular disease (CVD) and type 2 diabetes, the PAQ-C was used to classify children's physical activity levels. Body composition, physical inactivity, and cardiorespiratory fitness were also investigated in this study to determine their prediction ability. Obese children ($N = 83$) ages 6 to 12 years of age from Edmonton and the surrounding area participated in this study.

Children were tested for CVD and type 2 diabetes risk factors. Dyslipidemia, insulin resistance, and elevated blood pressure were measured for; as well, assessments of height, weight, BMI, skinfolds, and total body and regional body composition (as assessed by Dexa scans) were completed. The sum of 5 skinfolds was used to classify children as obese. The children completed the PAQ-C and a physical maturation questionnaire, and the parents filled out a questionnaire classifying their social economic status. Physical inactivity was estimated from the previous 7 days of self-reported time spent watching television and playing video/computer games. Ethnic background, CVD, and type 2 diabetes histories were reported by parents. The children completed a

cardiovascular test, blood samples were taken, and blood pressure, insulin levels, and glucose levels were measured. Children were categorized as either low health risk (LHR) or high health risk (HHR). Those children with 1 or more risk factor for CVD or type 2 diabetes were categorized as HHR, whereas no risk factors produced a LHR status. Risk factors for CVD and type 2 diabetes included dyslipidemia, insulin resistance, and elevated blood pressure.

Fifty-three children were categorized as HHR and 30 children were categorized as LHR. Together, 24/83 children had 2 or more risk factors. The LHR children and the HHR children had almost identical activity levels as assessed by the PAQ-C ($M = 3.01$, $SD = 0.65$ and $M = 3.00$, $SD = 0.66$; respectively). Significant predictors of an HHR classification were central body fat mass and the sum of 5 skinfolds. LHR children and HHR children differed significantly ($p = 0.05$) in mean total body lean mass, mean total body fat mass, and mean central body fat mass. Physical activity along with other lifestyle variables did not predict metabolic risk for CVD and type 2 diabetes. Central body fat mass was the strongest predictor of an HHR categorization.

Carter, L. M., Whiting, S. J., Drinkwater, D. T., Zello, G. A., Faulkner, R. A., & Bailey, D. A. (2001). Self-reported calcium intake and bone mineral content in children and adolescents. Journal *of the American College of Nutrition, 20,* 502-509.

The physical activity levels of children and adolescents were assessed by the PAQ-C to determine whether or not physical activity has a relationship to bone mineral density. Two hundred and twenty-seven children from the Saskatchewan pediatric bone mineral accrual study were assessed in the fall of 1993. Additional measurements administered were DEXA scans to assess bone mineral, two to four 24-hour recalls monitoring dietary intake, maturity ratings, anthropomorphic measures such as height, weight, and adiposity. High school students were given a modified PAQ-C that omitted the recess item (i.e., PAQ-A).

Generally, in comparison to females, the males had higher activity scores. Males had a mean physical activity score of 3.1 ($SD = 0.6$, $n = 107$) and the females mean activity score was 2.7 ($SD = 0.6$, $n = 117$). For both males and females, there were no significant correlations between activity scores and lumbar spine bone mineral content or total body bone mineral content. Using a multiple linear regression model of lumbar spine bone mineral content in males and females, the activity score was a significant predictor of lumbar spine bone mineral content only for females.

Crocker, P. R. E., Eklund, R. C., & Kowalski, K. C. (2000). Children's physical activity and physical self-perceptions. *Journal of Sports Sciences, 18,* 383-394.

The PAQ-C and the Physical Self-Perception Profile (PSPP) were used to study the relationship between children's physical activity levels and physical self-perceptions. Two hundred and twenty boys and 246 girls were recruited. The children were in grades 5 to 8 and were selected from five different schools.

The PAQ-C scores differed significantly between the boys and the girls ($p < 0.008$). The mean activity scores for boys, girls, and boys and girls combined were 3.21(SD = 0.67), 2.95 (SD = 0.64), and 3.07 (SD = 0.67) respectively. A multivariate analysis of variance of the PAQ-C activity checklist displayed that the frequency of reporting certain activities differed between boys and girls (Wilks' lambda = 0.638, $F_{22, 451} = 11.63$, $p <$ 0.001). However, the effect sizes for most activity differences reported were small. The authors suggested caution when interpreting the differences between the boys' and the girls' activities because the PAQ-C questionnaire is based only on the previous 7 days.

Children's physical activity levels were significantly correlated with self-perceptions of sport competence, body attractiveness, physical conditioning, physical strength, and general physical self-worth (r = 0.26-0.47 for girls and r = 0.28-0.47 for boys, $p < 0.05$). The physical self-perception model with pathways from physical condition and sport competence to the PAQ-C predicted 27 to 29 percent of the variance in boys' and girls' activity scores.

Crocker, P. R. E., Holowachuk, D. R., & Kowalski, K. C. (2001). Feasibility of using the Tritrac motion sensor over a 7-day trial with older children. ***Pediatric Exercise Science, 13,*** **70-81.**

To assess the feasibility of the Tritrac-R3D activity monitor (Tritrac), the PAQ-C along with other physical activity measures were compared to the Tritrac. Seventy-nine children in grades 4 to 8 participated. The final sample consisted of 34 girls and 27 boys who were predominately Caucasian and lived in a middle class area. The Tritrac and the Caltrac personal activity computer (Caltrac) were used by the children. The children also completed the PAQ-C and the 7-day physical activity recall interview (PAR). They wore the Tritrac and Caltrac monitors for 7 consecutive days.

The PAQ-C was significantly correlated with the PAR summary score (r = 0.39); however, the PAQ-C was not related to the Tritrac (r = 0.13), and the Tritrac was related to the Caltrac (r = 0.86). One explanation as to why the Tritrac was not correlated with the PAQ-C may be due to adherence issues with the Tritrac. Problems with the Tritrac included mechanical problems, social embarrassment, discomfort, and forgetting to wear the device. Of the 79 students that participated in this study, 22 students did not reach 56 hours of Tritrac data per week. The adherence problems with the Tritrac may have distorted the children's normal activity patterns, which may explain the low correlations between subjective and objective measures. The results of this study suggest that the Tritrac may not be feasible for studies of longer duration assessing general levels of physical activity in children.

Crocker, P. R. E., Sabiston, C., Forrestor, S., Kowalski, N. P., Kowalski, K. C., & McDonough, M. (2003). Predicting change in physical activity, dietary restraint, and physique anxiety in adolescent girls. ***Canadian Journal of Public Health, 94,*** **332-337.**

The PAQ-A was used in this two-year study to measure adolescent females' physical activity levels. The girls' body mass index (BMI), global self-esteem (GSE), physical self-perceptions, social physique anxiety (SPA), and dietary restraint were also examined. Of most importance were the changes in these constructs and the relationships that followed.

Six hundred and thirty-one female adolescents (ages 15-16) participated. The girls were enrolled in schools in Saskatoon and the surrounding area. They completed the Physical Self-Perceptions Profile, the Dutch Eating Behavior Questionnaire-Restrained Eating (DEBQ-R), the PAQ-A, and the Social Physique Anxiety Scale during class time. BMIs were also calculated (range = 14.8-36.9) based on self-reported height and weight.

The adolescent females' physical activity scores decreased (year 1 $M = 2.65$, $SD = 0.59$; year 2 $M = 2.40$, $SD = 0.55$; $p < 0.05$). Cross-sectional analysis for the first year revealed that the PAQ-A was significantly correlated ($p < 0.05$) to GSE, BMI, and physical self-perceptions of physical self-worth (PSW), body appearance, conditioning, sport, and strength (range = 0.12-0.51). For year 2, the PAQ-A was correlated to the same variables as year 1 (range = -0.12-0.58), except for BMI (and also with the addition of SPA). Pearson product moment correlations displayed that a change in physical activity was related to a change in each self-perception (excluding BMI), and physical conditioning was found to be the strongest predictor of physical activity change.

Ernst, M. P., & Pangrazi, R. P. (1999). Effects of a physical activity program on children's activity levels and attraction to physical activity. ***Pediatric Exercise Science, 11,*** **393-405.**

The impact of both an activity intervention (termed P.L.A.Y.) and a modified P.L.A.Y. placebo intervention on elementary school children was partially assessed by the PAQ-C. The PAQ-C was used to monitor activity levels from the start of the study to following the intervention. The teachers from 5 elementary schools ($N = 28$) and their students participated. Each class consisted of approximately 25 students. The students were in either grades 4, 5, or 6 and the ratio of boys to girls was approximately balanced. The majority of teachers and students were Caucasian and were of middle economic status. The children were categorized as either high or low activity levels from their pre-test PAQ-C scores. The PAQ-C and the Children's Attraction to Physical Activity (CAPA) scale were administered at the same time.

The children were randomly assigned to either the intervention group or the placebo group before the study began. The P.L.A.Y. intervention and the P.L.A.Y. placebo intervention were 12 weeks in length and consisted of 2 steps. The first step (a duration of 4 weeks) included a 15-minute activity break during the school day. During these weeks, the P.L.A.Y intervention group was encouraged to participate in activity and their teachers taught a new activity game each day (total of 15 games), whereas the placebo group was not encouraged to be active and no games were taught. The children were given a logbook to complete step 2 (8 weeks in duration). The P.L.A.Y intervention group logged their activity hours from the previous day, and the P.L.A.Y. placebo group recorded the number of hours spent watching television. At this step the 15-minute activity breaks were discontinued for both groups.

The intervention group consistently had significantly higher activity scores compared to the placebo group ($p < 0.05$). The intervention boys' and girls' mean baseline physical activity scores were 3.05 ($SD = 0.68$) and 2.89 ($SD = 0.54$) respectively, whereas the control boys' and girls' mean baseline physical activity scores were 3.01 ($SD = 0.72$) and 2.77 ($SD = 0.50$) respectively. The mean activity scores for the intervention boys' at the midpoint and at the end of the intervention were 3.40 ($SD = 0.57$) and 3.37 ($SD = 0.48$) respectively, whereas the placebo boys' mean activity scores were 2.93 ($SD = 0.66$) and 3.02 ($SD = 0.56$) respectively. The girls had similar results. The intervention girls' mean activity scores at the midpoint and at the endpoint were 3.16 ($SD = 0.43$) and 3.09 ($SD = 0.40$) respectively, whereas the placebo girls' mean scores were 2.80 ($SD = 0.52$) and 2.76 ($SD = 0.39$) respectively. The effect size for both the intervention boys and girls were significant at the midpoint and post-test (0.71 and 0.63 respectively for boys and 0.69 and 0.85 for girls). The P.L.A.Y. intervention children who were classified in the high activity group always had higher activity scores than the placebo group. The majority of children with high and low physical activity levels who participated in the P.L.A.Y intervention improved their physical activity status.

Gurd, B., & Klentrou, P. (2002). Physical and pubertal development in young male gymnasts. *Journal of Applied Physiology, 95,* 1011-1015.

This study measured adolescent boys' weekly energy expenditure with the help of the PAQ-A. The boys were gymnasts and had intense training programs. Their physical growth and sexual maturation were investigated for training effects.

Twenty-one elite male gymnasts and 24 controls (enrolled in a martial arts school) were recruited to participate. Each gymnast was participating in at least 15 hours of training per week, whereas the controls completed 2 hours or less per week (M age = 13.3 years, M age = 13.5 years; respectively). Measurements of height, weight, pubertal maturation, salivary testosterone, and relative body fat (as assessed by bioelectrical impedance) were taken once before the adolescents' regular training. As well, the adolescents reported the number of training sessions and training hours per week. The PAQ-A assisted the measurement of weekly energy expenditure [metabolic equivalents (MET)/wk].

The PAQ-A general physical activity scores were not reported. Energy expenditure (MET/wk), training sessions/wk, and training duration (h/wk) made up the males' physical activity characteristics. The elite male gymnasts had significantly ($p \leq 0.05$) greater energy expenditures, training duration, and training sessions per week compared to the controls. Training variables and energy expenditure were significantly ($p \leq 0.05$) correlated to relative body fat but not to physical growth or sexual maturation.

Heinonen, A., McKay, H. A., Whittall, K. P., Forster, B. B., & Khan, K. M. (2001). Muscle cross-sectional area is associated with specific site of bone in prepubertal girls: A quantitative magnetic resonance imaging study. *Bone, 29,* 388-392.

In this study, a modified PAQ-C was used to estimate the duration (hours per week) of loaded physical activity children performed. The possible relationship between muscle cross-sectional area and total cortical bone area in prepubertal and early pubertal girls was of greatest importance. Specifically, this study investigated whether or not a region-specific relationship existed.

Prepubertal and early pubertal girls (N = 17) who were participating in regular physical education classes; of tanner stage I-II; had no metabolic disorders and musculoskeletal complications; and were not taking medications that would influence bones, balance, or strength participated in this study. The girls attended elementary school and were between the ages of 9 to 11 years. The children's height and weight were measured, and a maturity rating was established. Calcium intake and health history were also assessed. Muscle and bone measurements were measured by an MRI, lower limb explosive performance capacity was assessed, and drop jump and side-to-side jump ground reaction forces were evaluated. The duration (h/week) of loaded physical activity was estimated from the PAQ-C's after school and during school items.

The PAQ-C scores were not reported; however, all girls were reported to be moderately active. The mean time that children spent in loaded physical activity was 5 hours/week (SD = 3.5).

MacKelvie, K. J., McKay, H. A., Khan, K. M., & Crocker, P. R. E. (2001a). Lifestyle risk factors for osteoporosis in Asian and Caucasian girls. ***Medicine and Science in Sports and Exercise, 33*****, 1818-1824.**

The PAQ-C was used to assess the level of children's participation in loaded physical activity. MacKelvie, McKay, Khan, and Crocker (2001) studied 191 young girls' (9-12 years of age) bone mineral content and associated lifestyle factors. The girls were classified as either Tanner breast stage I or Tanner breast stage II. There were 56 Asian girls and 75 Caucasian girls from 14 schools outside of Vancouver in the Richmond school district. The girls were classified into ethnic groups and the following measurements were taken: maturity ratings, bone mineral measures assessed by DEXA scans, height and weight measurements, calcium intake assessed by a food frequency questionnaire, and physical activity participation assessed by a modified PAQ-C. The modified PAQ-C included a measurement of time (h x wk^{-1}) spent in loaded physical activities (impact > walking) and a record of how often the child participated in evening organized sports and/or activity lessons per week.

The Tanner I Asian girls participated significantly less in general physical activity (p < 0.05) and had significantly lower calcium intake (p < 0.001) than the Tanner I Caucasian girls. The Tanner I Asian girls' mean scores for general physical activity, sport nights per week, and time spent in loaded physical activity (hr/wk) were 2.7(SD = 0.5), 1.0 (SD = 1.3), and 4.2 (SD = 4.0) respectively, whereas the Tanner I Caucasian girls' mean scores were 3.0 (SD = 0.6), 1.5 (SD = 1.7), and 4.6 (SD = 3.5) respectively. The Tanner I Caucasian girls spent 66% of their time in sport outside of school, whereas the Tanner I Asian girls spent 40% of their time in extracurricular sports. The Tanner I Caucasian girls contributed 9% more of their time in loaded physical activity per week compared to Tanner I Asian girls.

The Tanner II Caucasian girls had significantly higher loaded physical activity participation ($p < 0.01$), sports nights per week ($p < 0.05$), and calcium intake ($p < 0.001$) than the Tanner II Asian girls. The Tanner II Caucasian girls' mean scores for general physical activity, sport nights per week, and time spent in loaded physical activity (hr/wk) were 2.9 (SD = 0.6), 2.5 (SD = 1.8), and 5.9 (SD = 4.1) respectively. In comparison to the Tanner II Caucasian girls, the Tanner II Asian girls' mean score for general physical activity was similar (2.7, SD = 0.6), lower for sport nights per week (1.3, SD = 2.0), and lower for time spent in loaded physical activity (3.3, SD = 2.7). The Tanner II Caucasian girls had approximately 41% higher calcium intake, more participation in out-of-school sports per week, and 44% more time in loaded activity.

MacKelvie, K. J., McKay, H. A., Khan, K. M., & Crocker, P. R. E. (2001b). A school-based exercise intervention augments bone mineral accrual in early pubertal girls. ***The Journal of Pediatrics, 139*****, 501-508.**

The PAQ-C was used to examine whether or not physical education exercise interventions may be beneficial for bone health in prepubertal and early pubertal girls. Fourteen mixed ethnic schools from the Richmond school district outside of Vancouver participated in a school-based physical activity intervention. The schools were randomly assigned to either the control or the intervention group. This study focused on the girls who participated (26 prepubertal (PRE) control, 44 PRE intervention, 64 Early puberty (Early) control, and 43 Early intervention). The girls underwent the following measurements: maturity ratings, bone mineral measurements assessed by DEXA scans, height and weight, calcium intake assessed by a food frequency questionnaire, and physical activity participation assessed by a modified PAQ-C. The modified PAQ-C included a measurement of time spent in loaded activities and in common sports, as well as a measurement of how often the child participated in evening organized sports. The children in the intervention group participated 3 times per week in a 10-12 minute, high impact, weight bearing training program where the intensity of the program was increased progressively.

The intervention group and the control group for both prepubertal and early puberty girls had a similar mean calcium intake, general physical activity level (PRE control = 2.80 [0.47], PRE intervention = 2.96 [0.52], Early control = 2.90 [0.52], and Early intervention = 2.90 [0.51]), loaded physical activity (PRE control = 4.5 [3.2], PRE intervention = 4.9 [3.0], Early control = 5.7 [3.7], and Early intervention = 5.7 [4.1]), and extracurricular sport participation per week (PRE control = 1.2 [1.2], PRE intervention = 1.2 [1.4], Early control = 1.7 [1.5], and Early intervention = 1.6 [1.4]). Compared to the Early girls in the control group, the Early girls who participated in the intervention accumulated 1.5% to 3.1% more bone at the lumbar spine and femoral neck ($p < 0.05$).

MacKelvie, K. J., McKay, H. A., Petit, M. A., Moran, O., & Khan, K. M. (2002). Bone mineral response to a 7-month randomized controlled, school-based jumping intervention in 121 prepubertal boys: Associations with ethnicity and body mass index. ***Journal of Bone and Mineral Research, 17,*** **834-844.**

Prepubertal boys' general physical activity levels were determined by the PAQ-C. Modifications were made to the PAQ-C so that the researchers could estimate the children's time spent in loaded activity and common sports. Of most interest to this study were the bone mineral changes in prepubertal boys after a 7-month jumping intervention. These changes were then compared to the control boys' bone mineral changes.

Fourteen schools outside of the Vancouver area volunteered to participate after being recruited. Three hundred and eighty-three children (ages 8.8-11.7 years) participated. The children were categorized as either Asian or Caucasian. If a child was taking medications or had a medical complication that could influence physical activity or bone development they were excluded from the study. A health history questionnaire assessing the children's health was completed by the parents. The following measurements were also taken: maturity rating, bone mineral, height, weight, calcium, and physical activity. A modified PAQ-C estimated the time (h/week) the boys spent in out-of-school sports and loaded activities (impact > walking). The mean general physical activity scores reported in this study are an average of three assessments by the PAQ-C during the school year (baseline, winter, and a final measurement).

The intervention group participated in a school-based jumping exercise intervention 3 times per week (2 physical education class times and one supervised session). The exercise intervention included high-impact, weight-bearing activity for 10 to 12 minutes. The program's difficulty and loading progressed throughout the school year.

There was no change in prepubertal boys' physical activity scores and loaded activity time for control, intervention, Asian, and Caucasian groups over the 7 months. The baseline mean physical activity score for the control boys was 3.1 ($SD = 0.5$) and 3.1 ($SD = 0.6$) for the intervention group. Asian and Caucasian control groups with an average body mass index (BMI) had mean baseline physical activity scores of 3.0 ($SD = 0.4$) and 3.0 ($SD = 0.5$) respectively. The intervention Asian and Caucasian groups with an average BMI had mean baseline physical activity scores of 3.0 ($SD = 0.6$) and 3.1 ($SD = 0.6$). The mixed ethnicity control boys had a mean baseline PAQ-C score of 3.1 ($SD = 0.6$). The mean was 3.0 ($SD = 0.4$) for the mixed ethnicity intervention group. At several different sites, the intervention boys (Asian and Caucasian) with low or average BMI gained significantly ($p < 0.01 - p < 0.05$) more bone mineral accrual than their controls. No changes were found in prepubertal intervention and control boys with a high BMI over the 7 months. The jumping intervention was reported as successful in increasing bone mineral accrual in Asian and Caucasian prepubertal boys with a low or average BMI.

MacKelvie, K. J., Petit, M. A., Khan, K. M., Beck, T. J., & McKay, H. A. (2004). Bone mass and structure are enhanced following a 2-year randomized controlled trial of exercise in prepubertal boys. *Bone, 34,* 755-764.

Prepubertal boys' general physical activity levels were determined by the PAQ-C. Modifications were made to the PAQ-C so that the researchers could estimate the children's time spent in loaded activity and common sports. Of most interest to this study were the prepubertal boys' changes in proximal femur bone geometry and strength after a

2-year loaded physical activity intervention. These results were compared to the control boys' changes. As well, the intervention boys' and the control boys' total body, proximal femur, and lumbar spine bone mineral content and bone area changes were of importance.

Fourteen schools outside of the Vancouver area volunteered to participate after being recruited. All schools were randomly assigned to either the intervention or the control groups (intervention = 7 and control = 7). Thirty-one intervention boys and 33 control boys participated. A health history questionnaire assessing the children's health was completed by the parents. The following measurements were also taken at baseline, 8 months, 12 months, and 20 months: maturity rating, bone mineral content, bone area, height, weight, calcium intake, leg length, long jump, vertical jump, and physical activity. Structural and geometric characteristics of the proximal femur were assessed by a Hologic Inc. QDR-4500 bone densitometer. These measurements were analyzed using hip structural analysis. A modified PAQ-C assessed the number of nights the boys spent in common sports per week and the time spent in loaded activities (h/week). The mean general physical activity scores reported in this study are an average of 6 assessments by the PAQ-C over the 2 school years (fall, winter, and spring completion).

The intervention group participated in a school-based jumping exercise intervention. The intervention included 3 sessions per week with high-impact, weight-bearing activity for 10 to 12 minutes. The program's difficulty and intensity progressed throughout the 2 school years.

The PAQ-C average 20-month physical activity scores were similar between the intervention and control boys, $M = 3.2$ ($SD = 0.6$) and $M = 3.1$ ($SD = 0.5$) respectively. Further, the scores for average number of sports nights per week and average time spent in loaded activity were similar between the two groups.

Mahon, A. D., Anderson, C. S., Hipp, M. J., & Hunt, K. A. (2003). Heart rate recovery from submaximal exercise in boys and girls. ***Medicine and Science in Sports and Exercise, 35,*** **2093-2097.**

The PAQ-C was used to assess 11 boys' and 10 girls' physical activity levels. This study was interested in their heart rate (HR) changes that occurred after two separate submaximal exercise sessions.

The children's pubertal status, height, weight, and 6 skinfolds were measured. The parents completed the PAQ-C (with help from their children). To measure peak VO_2, a graded cycle ergometer test was completed by the children on two different sessions. During these sessions resting HR, exercise HR, and postexercise HR were measured.

The boys and girls had similar physical activity levels ($M = 3.1$, $SD = 0.6$; $M = 3.0$, $SD = 0.4$; respectively). In this study, peak VO_2 was less effective in accounting for variations in postexercise HR in comparison to resting HR.

McKay, H. A., Petit, M. A., Khan, K. M., & Schutz, R. W. (2000). Lifestyle determinants of bone mineral: A comparison between prepubertal Asian- and Caucasian-Canadian boys and girls. ***Calcified Tissue International, 66,*** **320-324.**

McKay, Petit, Khan, and Schutz (2000) compared Caucasian and Asian children's lifestyle factors that could affect bone mineral content. Ten Canadian multi-ethnic schools participated. Fifty-eight prepubertal Asian children and 110 prepubertal Caucasian children (mean age 8.9) were assessed. The children received Dexa scans of the proximal femur, lumbar spine, and total body. The following measurements were taken: stretch statures as assessed by a wall stadiometer, maturity ratings, and weight. The children completed a food frequency questionnaire to classify calcium intake and the PAQ-C to classify physical activity levels. The scoring of the PAQ-C was modified. The original PAQ-C provides an activity rating from 1 to 5 for each child (where a 1 represents a low activity level and a 5 represents a high activity level). In this study, the physical activity score was calculated from the amount of loaded physical activity that was recalled by the children in the prior week and was not scored on a 5-point scale. Additionally, the children were asked if the activity reported for the previous week was consistent with their usual activity participation.

The PAQ-C activity scores demonstrated the differences in Asian and Caucasian children's physical activity levels. Overall, Asian children had a significantly lower calcium intake and physical activity participation than the Caucasian children (70.2 [SD = 19.2] vs. 82.0 [SD = 16.5] respectively; $p < 0.001$). The mean activity score for Asian children was 67.4 (SD = 16.6) for boys and 73.3 (SD = 21.5) for girls, whereas the mean activity score for Caucasian children was 87.8 (SD = 17.1) for boys and 76.0 (SD = 13.5) for girls. Only 14% of Asian boys reported participating in sport, compared to 73% of Caucasian boys reporting involvement in organized sport. Bone mineral free lean mass, sex, and physical activity were significant predictors of areal bone mineral density at the femoral neck and the total proximal femur.

McKay, H. A., Petit, M. A., Schutz, R. W., Prior, J. C., Barr, S. I., & Khan, K. M. (2000). Augmented trochanteric bone mineral density after modified physical education classes: A randomized school-based exercise intervention study in prepubescent and early pubescent children. ***The Journal of Pediatrics, 136,*** **156-162.**

The PAQ-C was used to classify children's activity levels in order to determine the impact of an 8-month exercise intervention on children's areal bone mineral density. One hundred and forty-four children in grades 3 and 4 from 10 different elementary schools were assessed. Forty-nine children were Asian and 95 children were Caucasian. The schools were randomly placed into either the intervention or the control group. The intervention group participated in a loaded activity physical education intervention, whereas the control group participated in their usual physical education class. The intervention was performed 3 times per week and included 10 minutes of loaded activity, 10 tuck jumps at the beginning of class, and 10 to 30 minutes of total activity. The intensity of the intervention progressed throughout the 8 months. The children received Dexa scans and anthropometric measurements. They also completed calcium intake questionnaires, a healthy history form, a self-assessment of maturity, and the PAQ-C. The

PAQ-C was modified in order to focus on the children's loaded activity levels and the final activity score was not based on a 5-point scale.

According to the baseline measurements of the PAQ-C, the children in the control group had significantly higher physical activity levels than the intervention group (control = 86.0, exercise = 80.5, $p = 0.002$). The children who participated in the exercise intervention had significantly greater gains in femoral trochanteric areal bone mineral density than the control group (4.4% vs. 3.2 %, $p = 0.05$). Interestingly, the control group had greater activity levels compared to the intervention group at the 8-month final assessment.

Muratova, V. N., Islam, S. S., Demerath, E. W., Minor, V. E., & Neal, W. A. (2001). Cholesterol screening among children and their parents. ***Preventive Medicine, 33,*** **1-6.**

The PAQ-C was used to classify children's physical activity levels in a school-based cholesterol-screening program. Seven hundred and nine children in the fifth grade from rural Appalachian countries participated. The children were comprised of 326 boys and 383 girls (mean age 10.8) where the majority of children were Caucasian. The cholesterol-screening program was comprised of three phases: screening, diagnosis, and intervention. The children's nonfasting finger-stick total blood cholesterol, high-density lipoprotein cholesterol, anthropomorphic measures, and dietary history were assessed in the screening phase. Additionally, physical activity levels were assessed by the PAQ-C, parental smoking was measured by a questionnaire completed by the parents, and anxiety levels were assessed on a 5-point Likert scale. The children's mean physical activity score was 2.89 ($N = 707$, $SD = 0.64$). The PAQ-C physical activity score displayed a relationship with diastolic blood pressure; however, the magnitude of the relationship was weak ($r = -0.07$).

Paxton, R. J., Estabrooks, P. A., & Dzewaltowski, D. (2004). Attraction to physical activity mediates the relationship between perceived competence and physical activity in youth. ***Research Quarterly for Exercise and Sport, 75,*** **107-111.**

To determine children's physical activity levels, the PAQ-C was used. Relationships among children's physical activity, perceived physical competence, and attractions to physical activity were investigated. Further, this study examined whether or not attraction to physical activity mediates the relationship between perceived physical competence and physical activity.

The children of four 4-H youth development clubs were asked to participate. Sixty-three Caucasian children (ages 9-14 years) from the entire sample of 94 children provided parental consent to participate. Thirty-four percent of children were boys. A modified Perceived Physical Competence Scale for children, The Children's Attraction to Physical Activity Scale (CAPAS), and the PAQ-C were completed during the winter (*Note. In this study the PAQ-C was called the PAQ-OC*).

The children had low physical activity levels (M = 1.83, SD = 0.64). Significant ($p < 0.01$) bivariate correlations were found between physical activity and attraction to physical activity (r = .45) and perceived physical competence (r = .34). Further, attraction to physical activity mediated the relationship between perceived physical competence and physical activity.

Petit, M. A., McKay, H. A., MacKelvie, K. J., Heinonen, A., Khan, K. M., & Beck, T. J. (2002). A randomized school-based jumping intervention confers site and maturity-specific benefits on bone structural properties in girls: A hip structural analysis study. ***Journal of Bone and Mineral Research, 17,*** **363-372.**

Prepubertal and early pubertal girls' general physical activity levels were assessed by the PAQ-C. This study focused on what geometric and structural bone changes occurred in the girls as a result of a school-based jumping exercise intervention.

Fourteen schools outside of the Vancouver area volunteered to participate after being recruited. One hundred and seventy-seven girls (ages 9-12 years) volunteered to participate. The schools were randomly assigned to either the intervention or control group. The girls' stretch statures, sitting heights, weights, calcium intakes, maturity ratings, and physical activity levels were measured. A modified PAQ-C was used so that a measure of loaded activity (impact > walking, h/week) and the number of nights the girls' participated in organized sport per week could be calculated. The girls completed the PAQ-C three times during the year.

The loaded exercise intervention included high-impact, 10 to 12 minutes sessions, and a variety of jumping activities. Two sessions per week were held during the girl's physical education class, while the third session was completed in class time on a different day. The difficulty and the intensity of the intervention progressively increased throughout the year.

The PAQ-C scores were not included. However, this study reported that the baseline measurements of physical activity for the intervention and control group showed no differences. Further, the abstract reported that physical activity levels were similar for both groups over the 7 months.

Rosendo da Silva, R. C., & Malina, R. M. (2000). Level of physical activity in adolescents from Niteroi, Rio de Janeiro, Brazil. ***Cadernos-de-saude-publica-Ministerio-da-Saude,-Fundacao-Oswaldo-Cruz,-Escola-Nacional-de-Saude-Publica, 16,*** **1091-1097.**

Rosendo da Silva and Malina (2000) created a Portuguese version of the PAQ-C to assess general physical activity levels in Brazilian children and adolescents. The manuscript and the PAQ-C were in Portuguese; therefore, this study summary is based on its English written abstract and observations made from the Portuguese PAQ-C.

Rosendo da Silva and Malina (2000) focused on Brazilian adolescents' (N = 325) activity levels. Anthropomorphic measures, hours of television viewing per day, and general activity levels were measured. The males' mean PAQ-C activity score was 2.3

(SD = 0.6, $p < 0.01$) and the females' mean activity score was 2.0 (SD = 0.6, $p < 0.01$). The most frequently chosen activity was soccer for males and soccer and walking for females. A high percentage of children were classified as inactive from their PAQ-C scores (85% of males and 94% of females were inactive). The authors of this study were concerned with the high percentage of inactive children because these children may be more likely to be inactive adults. Interestingly, the mean activity scores for the Brazilian females and males were slightly lower than the mean activity scores found in the English PAQ-C validation reliability studies (see Chapter 2 for the English PAQ-C scores).

The Portuguese PAQ-C has content and scoring differences compared to the English PAQ-C. The Portuguese PAQ-C has additional items and includes a television item quantifying the number of hours children and adolescents watch television per day (this item is not included in the final activity score). Both questionnaires have an activity checklist item; however, the number of activity choices differ. The Portuguese version of the PAQ-C has 15 activity choices, whereas the original PAQ-C has 23 activity choices. Therefore, researchers should be cautious in interpreting the mean value of the activity checklist item because the Portuguese activity checklist item may be prone to producing a higher or lower activity score than the original PAQ-C.

Rourke, K. M., Brehm, B. J., Cassell, C., & Sethuraman, G. (2003). Effect of weight change on bone mass in female adolescents. ***Journal of the American Dietetic Association, 103,*** **369-372.**

The activity levels of obese female adolescents were assessed by the PAQ-C. This study was interested in obese adolescents' changes in bone mineral density and bone mineral content as a result of participating in a weight reduction program. Ninety-two obese girls (mean age = 11) were recruited through newspaper advertisements and recommendations by pediatricians. The children received Dexa scans, anthropomorphic measures, and a body composition measure. They also completed the PAQ-C. The authors used the PAQ-C data to calculate the adolescent's energy expenditure.

However, the physical activity data were not presented, although the authors stated that the activity levels of the obese adolescent girls were similar to those of a normal weight adolescent population. Also, physical activity levels were not significant predictors of bone loss or gain.

Sirard, J. R., & Pate, R. P. (2001). Physical activity assessment in children and adolescents. ***Sports Medicine, 31,*** **439-454.**

The strengths, limitations, and validity of subjective and objective measures of physical activity in children and adolescents were reviewed. This study reviewed criterion standards (direct observation, doubly labelled water, and indirect calorimetry), secondary measures (heart rate, pedometers, and accelerometers), and subjective measures (self-report, interview, proxy-report, and diary).

Self-report questionnaires' time frames, participants, reliability, criterion measure, and validity were reported in a table. The PAQ-C was one of the measures listed, but was not discussed in the text. Limitation in subjective measures discussed included children's

recall ability and their struggle to recall intensity and type of activity. The authors suggested that survey measures be validated against strong standards of physical activity assessment.

Thompson, A. M., Baxter-Jones, A. D. G., Mirwald, R. L., & Bailey, D. A. (2003). Comparison of physical activity in male and female children: Does maturation matter? ***Medicine and Science In Sports and Exercise, 35,*** **1684-1690.**

In this longitudinal study (from years 1991-1997), children's general physical activity levels were assessed using the PAQ-C. The boys' and girls' biological ages (chronological age at peak height velocity subtract chronological age at time of measurement) were considered during the analysis of their physical activity levels.

Seventy boys and 68 girls from the Saskatchewan bone mineral accrual study participated. Data collected when the children were ages 9 to 18 years were analyzed. The children's chronological age, height, body mass, biological age, and physical activity levels were measured. During the first 3 years of the study, the children completed the PAQ-C 3 times per year. Every year after, the PAQ-C was completed twice per year. The PAQ-C assessment scores for one year were averaged to make up a yearly summary activity score. The PAQ-A (the PAQ-C omitting the recess item) assessed high school students' physical activity levels.

When physical activity scores were analyzed by chronological age, significant ($p < 0.05$) sex differences were found between the ages of 10 to 16 years. The boys had higher activity scores compared to the girls. Further, boys' and girls' physical activity levels decreased as chronological and biological age increased. No sex differences were found (with an exception at 3 years before peak height velocity) in physical activity levels when biological age was controlled for ($p < 0.05$).

Welk, G. J., Corbin, C. B., & Dale, D. (2000). Measurement issues in the assessment of physical activity in children. ***Research Quarterly for Exercise and Sport, 71,*** **59-73.**

The PAQ-C's strengths and limitations were mentioned in this study's review of physical activity assessment. However, the PAQ-C was not included in the comparison of self-report measures. It was mentioned as an assessment tool that measures children's general physical activity levels. The authors noted that the PAQ-C's general measurement is beneficial for studies that do not need estimates of time or amounts of activity. Further, the PAQ-C was recommended as an instrument that may have the ability to differentiate between active and inactive children. The authors reported that the PAQ-C does not provide a measurement of duration, frequency, and intensity.

Welk, G. J., & Wood, K. (2000). Physical activity assessments in physical education: A practical review of instruments and their use in the curriculum. ***Journal of Physical Education, Recreation and Dance, 71,*** **30-40.**

Welk & Wood (2000) reviewed a variety of physical activity measures, and concluded that the PAQ-C, along with other self-report measures have the potential to be used in physical education evaluation. This study reported two limitations of the PAQ-C. First, each item is scored on a 5-point scale and the final activity score is a composite score made up of 9 items. Giving equal weight to each item was viewed as a possible limitation because the items have different time periods for possible activity. Second, the PAQ-C does not provide an estimate of caloric expenditure. The PAQ-C was recognized for its ability to differentiate children into either active or non-active categories.

Welk, G. J., Wood, K., & Morss, G. (2003). Parental influences on physical activity in children: An exploration of potential mechanisms. ***Pediatric Exercise Science, 15,*** **19-33.**

A parent's influence on their child's physical activity level was examined with the help of the PAQ-C. Children in grades 3 to 6 from three schools participated in this study. There were 994 students (505 boys and 489 girls) along with 536 parents. Only one parent from each family was asked to participate (82% were mothers, 17% fathers, and 1% were guardians). The children completed the PAQ-C and the Children's Physical Activity Correlates (CPAC) instrument during their physical education class. The CPAC consisted of an attraction to sport scale, a perceptions of competence scale, a parental influence scale, a parental role modelling scale, a parental support scale, a parental encouragement scale, a parental involvement scale, and a parental facilitation scale. The parents completed two physical activity measures at home, which assessed their physical activity level and categorised them as inactive, moderately active, or regularly active.

The boys had significantly higher activity levels than the girls, F (1,986) = 10.7, $p <$ 0.01. The mean activity score for boys was 3.29 (SD = 0.66), whereas the mean activity score for girls was 3.16 (SD = 0.62). The effect size was small for the difference between boys' and girls' activity scores (0.20).

The PAQ-C was moderately related to the attraction scale, perceptions of competence scale, and the parental influence scale (r = 0.38 to r = 0.52). Together, the parental support and parental role modelling variables predicted 19.7% of the variance in the PAQ-C scores, F (2,991) = 121.3, $p < 0.001$. The parents' activity levels (NOT assessed with the PAQ-C) were weakly correlated to their children's (r = 0.13 to r = 0.16).

4.2 Reference List of Studies that have Cited or Used the PAQ-C and/or the PAQ-A

The following list of references includes 4 categories of studies: (1) the PAQ-C and PAQ-A development, validation, and reliability studies, (2) studies that used the PAQ-C and/or the PAQ-A, (3) manuscripts that reviewed the PAQ-C, and (4) manuscripts that cited the original PAQ-C and/or PAQ-A validation studies.

The PAQ-C and PAQ-A development, validation, and reliability studies

Crocker, P. R. E., Bailey, D. A., Faulkner, R. A., Kowalski, K. C., & McGrath, R. (1997). Measuring general levels of physical activity: Preliminary evidence for the Physical Activity Questionnaire for Older Children. *Medicine and Science in Sports and Exercise, 29,* 1344-1349.

Kowalski, K. C., Crocker, P. R. E., & Faulkner, R. A. (1997). Validation of the Physical Activity Questionnaire for Older Children. *Pediatric Exercise Science, 9,* 174-186.

Kowalski, K. C., Crocker, P. R. E., & Kowalski, N. P. (1997). Convergent validity of the Physical Activity Questionnaire for Adolescents. *Pediatric Exercise Science, 9,* 342-352.

Used the PAQ-C and/or the PAQ-A

Bailey, D. A. (1997). The Saskatchewan pediatric bone mineral accrual study: Bone mineral acquisition during the growing years. *International Journal of Sports Medicine, 18,* S191-S194.

Bailey, D. A., McKay, H. A., Mirwald, R. L., Crocker, P. R. E., & Faulkner, R. A. (1999). A six-year longitudinal study of the relationship of physical activity to bone mineral accrual in growing children: The University of Saskatchewan bone mineral accrual study. *Journal of Bone and Mineral Research, 14*, 1672-1679.

Ball, G. D. C., Marshall, J. D., & McCargar, L. J. (2003). Fatness and fitness is obese children at low and high health risk. *Pediatric Exercise Science, 15*, 392-405.

Carter, L. M., Whiting, S. J., Drinkwater, D. T., Zello, G. A., Faulkner, R. A., & Bailey, D. A. (2001). Self-reported calcium intake and bone mineral content in children and adolescents. *Journal of the American College of Nutrition, 20,* 502-509.

Crocker, P. R. E., Eklund, R. C., & Kowalski, K. C. (2000). Children's physical activity and physical self-perceptions. *Journal of Sports Sciences, 18,* 383-394.

Crocker, P. R. E., Holowachuk, D. R., & Kowalski, K. C. (2001). Feasibility of using the Tritrac motion sensor over a 7-day trial with older children. *Pediatric Exercise Science, 13,* 70-81.

Crocker, P. R. E., Sabiston, C., Forrestor, S., Kowalski, N., Kowalski, K. C., & McDonough, M. (2003). Predicting change in physical activity, dietary restraint, and physique anxiety in adolescent girls. *Canadian Journal of Public Health, 94,* 332-337.

Ernst, M. P. & Pangrazi, R. P. (1999). Effects of a physical activity program on children's activity levels and attraction to physical activity. *Pediatric Exercise Science, 11,* 393-405.

Gurd, B., & Klentrou, P. (2002). Physical and pubertal development in young male gymnasts. *Journal of Applied Physiology, 95,* 1011-1015.

Heinonen, A, McKay, H. A., Whittall, K. P., Forster, B. B., & Khan, K. M. (2001). Muscle cross-sectional area is associated with specific site of bone in prepubertal girls: A quantitative magnetic resonance imaging study. *Bone, 29,* 388-392.

MacKelvie, K. J., Khan, K. M., Petit, M. A., Janssen, P. A., & McKay, H. A. (2003). A school-based exercise intervention elicits substantial bone health benefits: A 2-year randomised controlled trial in girls [Abstract]. *Pediatric, 112,* e447-e452.

MacKelvie, K. J., McKay, H. A., Khan, K. M., & Crocker, P. R. E. (2001a). Lifestyle risk factors for osteoporosis in Asian and Caucasian girls. *Medicine and Science in Sports and Exercise, 33*, 1818-1824.

MacKelvie, K. J., McKay, H. A., Khan, K. M., & Crocker, P. R. E. (2001b). A school-based exercise intervention augments bone mineral accrual in early pubertal girls. *The Journal of Pediatrics, 139*, 501-508.

MacKelvie, K. J., McKay, H. A., Petit, M. A., Moran, O., & Khan, K. M. (2002). Bone mineral response to a 7-month randomized controlled, school-based jumping intervention in 121 prepubertal boys: Associations with ethnicity and body mass index. *Journal of Bone and Mineral Research, 17,* 834-844.

MacKelvie, K. J., Petit, M. A., Khan, K. M., Beck, T. J., & McKay, H. A. (2004). Bone mass and structure are enhanced following a 2-year randomized controlled trial of exercise in prepubertal boys. *Bone, 34,* 755-764.

Mahon, A. D., Anderson, C. S., Hipp, M. J., & Hunt, K. A. (2003). Heart rate recovery from submaximal exercise in boys and girls. *Medicine and Science in Sports and Exercise, 35,* 2093-2097.

McKay, H. A., Petit, M. A., Khan, K. M., & Schutz, R. W. (2000). Lifestyle determinants of bone mineral: A comparison between prepubertal Asian- and Caucasian-Canadian boys and girls. *Calcified Tissue International, 66,* 320-324.

Muratova, V. N., Islam, S. S., Demerath, E. W., Minor, V. E., & Neal, W. A. (2001). Cholesterol screening among children and their parents. *Preventive Medicine, 33*, 1-6.

Paxton, R. J., Estabrooks, P. A., & Dzewaltowski, D. (2004). Attraction to physical activity mediates the relationship between perceived competence and physical activity in youth. *Research Quarterly for Exercise and Sport, 75,* 107-111.

Petit, M. A., McKay, H. A., MacKelvie, K. J., Heinonen, A., Khan, K. M., & Beck, T. J. (2002). A randomized school-based jumping intervention confers site and maturity-specific benefits on bone structural properties in girls: A hip structural analysis study. *Journal of Bone and Mineral Research, 17,* 363-372.

Rosendo da Silva, R. C. & Malina, R. M. (2000). Level of physical activity in adolescents from Niteroi, Rio de Janeiro, Brazil. *Cadernos-de-saude-publica-Ministerio-da-Saude, -Fundacao-Oswaldo-Cruz,-Escola-Nacional-de-Saude-Publica, 16,* 1091-1097.

Rourke, K. M., Brehm, B. J., Cassell, C., & Sethuraman, G. (2003). Effect of weight change on bone mass in female adolescents. *Journal of the American Dietetic Association, 103,* 369-372.

Thompson, A. M., Baxter-Jones, A. D. G., Mirwald, R. L., & Bailey, D. A. (2003). Comparison of physical activity in male and female children: Does maturation matter? *Medicine and Science In Sports and Exercise, 35,* 1684-1690.

Welk, G. J., Wood, K., & Morss, G. (2003). Parental influences on physical activity in children: An exploration of potential mechanisms. *Pediatric Exercise Science, 15,* 19-33.

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Appendix 24: Modified Borg Scale

Modified Borg Scale	
0	Nothing at all
0.5	Very very light
1	Very light
2	Light
3	Moderate
4	Something hard
5	Hard
6	
7	Very hard
8	
9	
10	Very very hard

Appendix 25: Instructional Video for YBT

https://youtube.com/shorts/FKtuNWQmyjU?feature=share

Appendix 26: Lower Limb Assessment Score (LLAS)

Pediatric rheumatology

Joint hypermobility: The use of a new assessment tool to measure lower limb hypermobility

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ABSTRACT

Objectives

The aim of the study was to compare the use of a new assessment tool for diagnosis of hypermobility in the lower limb to the Beighton score for generalised hypermobility.

Methods

Three groups of children were compared (n = 225) and included a "normal" population of 116 school children, a "possible hypermobile" group of 88 children attending a foot and gait clinic and a "known hypermobile" group of 21 children referred from a paediatrician or rheumatologist. The Beighton score was used to measure generalised hypermobility. The Lower Limb Assessment Score was used to measure hypermobility in the lower limbs.

Results

The Lower Limb Assessment Score was able to distinguish between the three groups of children better than the Beighton score. At a threshold of 5/9 indicating hypermobility, the Beighton score identified hypermobility in 34% of school children; the lower limb score identified hypermobility in 21% of school children after a threshold was identified. There was disagreement between the scores in school children where 26.7% of children appeared to have a positive Beighton score that was not accompanied by a positive lower limb score. In the "known hypermobile" group the Beighton score was positive in only 10% of children when the lower limb score was negative for hypermobility.

Conclusion

In this group of school children, the Beighton score appeared to over-diagnose hypermobility at the threshold of 5/9. Specific thresholds for diagnosis need to be set dependant on the age and ethnic group of the population being studied. The Lower Limb Assessment Score may be a useful score for health professionals specifically interested in lower limb hypermobility.

Key words

Children, hypermobility.

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Introduction

A joint is described as hypermobile if the range of motion is excessive when taking into account the age, sex and ethnic group of the subject (1). Hypermobility is more common in females than males and is known to reduce with age (2). Children therefore, are more flexible than adults but hypermobility can still be present when the joint range of motion is excessive. Hypermobile children suffer from similar joint complaints to adults (3) and the lower limb is affected most often (4,5). In one prospective study, up to 40% of hypermobile children developed symptoms of arthralgia in the course of one year (6).

Diagnosis of hypermobility is often made using the criteria developed by Beighton *et al.* (2). A Beighton score of 5/9 is most frequently used to define hypermobility but this arbitrary cut-off point may make the system insensitive, being inappropriate for different ages and different ethnic groups. The Beighton scale is also heavily weighted to measurement of the upper limb despite the majority of musculoskeletal complaints presenting in the lower limb. With this scoring system, only knee hyperextension is measured in the lower limb and thus subjects presenting with lower limb complaints may fail to be diagnosed if the hypermobility is confined to the lower limb.

Since many of the symptoms related to hypermobility present in the lower limbs, a valid and reliable tool that measures hypermobility in the lower limb would be useful to professionals who diagnose and treat lower limb pathology. This study tested the usefulness of a new assessment scale for lower limb hypermobility. The scale included movements of the joints occurring in several planes of motion rather than in just one direction. The movements included were typical of those used by health professionals such as rheumatologists, physiotherapists and podiatrists, when assessing the lower limb. The score takes around 15 minutes to complete when the examiner is familiar with the set criteria but some initial training may be required if the examiner is not familiar with the positioning used to isolate the joints, such as for knee rotation and midtarsal joint movements.

A comparison of the new scoring system to the Beighton score was made. Through comparison of the two scores, the study aimed to investigate whether the Beighton score was a reasonable measure of lower limb hypermobility and aimed to investigate whether a lower limb scoring system would fail to identify any children defined as hypermobile by the Beighton score. In order to validate the lower limb scoring system, it was compared to the Beighton score as well as with expert clinical opinion as to whether a child was hypermobile. Although most of the scoring systems are best used as a sliding scale of flexibility, the new assessment was tested for validity after a specific cut-off point was set.

Method

Ethical approval was granted by the local ethical committees.

Subjects

Three groups of children were included in the study and were chosen to represent differing levels of hypermobility. The first group consisted of children in primary classes at three London schools and were included to represent "normal". The second group included children attending a paediatric foot and gait clinic at a teaching hospital where a high prevalence of foot conditions related to hypermobility were seen but no child was diagnosed with hypermobility at the time of referral. This group were "possible hypermobile". The third group consisted of children known to be hypermobile, being diagnosed with hypermobility by a rheumatologist or paediatrician. This group were "known hypermobile". The method for assessment for this diagnosis was not known and might vary between referring sources. Usually a full joint examination had been carried out but if a specific scoring system was used, the score was rarely given in the referral.

In the school children, all children attending school on the days of the assessments were included if consent had been given by the parents, unless the child refused to consent on the day or met any of the exclusion criteria. Con-

sent forms were sent to all parents of children attending the primary schools and requested basic demographic information including child's age, sex and ethnic group. Incomplete forms were supplemented from school records. The children were assessed in a quiet classroom environment within their schools.
For the children attending the paediatrics clinic, all children attending for routine appointments over a 6 month period were assessed if informed consent was given by the parent and child and the exclusion criteria were not met. Consent was obtained from all children attending the clinic at the time of their clinical appointment and demographic information was collected at that time.
Children were excluded from the study when:

- A diagnosis of joint disease (i.e. Juvenile Idiopathic Arthritis) or connective tissue disease existed that might restrict normal joint motion.
- A current or previous orthopaedic condition (other than simple fracture) was present influencing normal movement (ie. talipes, tarsal coalition).
- The child had a diagnosis of neurological or neurodevelopmental disorders.
- The child was aged less that 5 years old or older than 16 years old.

Hypermobility assessment
All children were assessed using the Lower Limb Assessment Score providing a score to a maximum of 12 marks for each limb. The Beighton score was taken for all children providing a single score to a maximum 9 (see Appendix 1). Each child was also graded according to the clinical opinion of the assessor using three gradings (hypermobile, borderline hypermobility, normal) by undertaking a full lower limb examination prior to the application of any hypermobility score.
Two examiners undertook the assessments after inter-observer repeatability of the Lower Limb Assessment Score was tested using reliability analysis in a pilot study of 22 children (Intraclass correlation coefficient of 0.84 (95% CI = 0.62 to 0.93; F (1, 21) = 6.08, p = 0.001)).

Statistical analysis
The distributions of the data were initially examined for normality and parametric tests or non-parametric tests carried out based on the distributions found. P values less than 0.05 were considered statistically significant.

Results
The "normal" group included 116 children (66♀: 50♂) with a mean age of 7 years (SD 1.9 years). The "possible hypermobile" group included 88 children (46♀: 42♂) with a mean age of 9.89 years (SD 3.39 years). The "known hypermobile" group included 21 children (13♀: 8♂) of mean age 9.18 years (SD 3.55 years).

Association between the Beighton and LLAS scores with age
The clinic children were slightly older than the school children therefore associations with age for the Beighton score and the LLAS score were tested initially. Pearson correlation was used to test for an association. No association was found in any of the groups for either the Beighton with age ("normals": r= 0.12; "possible hypermobile": r=0.36; "known hypermobile": r = 0.2) or the LLAS with age ("normals": r = 0.21; "possible hypermobile": r = 0.46; "known hypermobile": r = 0.19).

Comparison of left to right sides for the Lower Limb Assessment Score
A paired t-test was used to test the null hypothesis of no significant difference occurring between left and right sides scores (n = 225) for the LLAS. The null hypothesis was accepted: no significant difference was found (p = 0.74). The LLAS was taken as a score out of 12 rather than a total score of 24 for the two limbs for further analysis.

Using the Beighton score to measure hypermobility in children
The numbers of children found to be hypermobile using the Beighton criteria were compared in the three groups. Hypermobility was defined as a score of 5/9 or greater (Beighton positive). Within the school children ("normals"), 39 children (34%, 95%CI = 27 to 41%) were found to be hypermobile using this criteria for hypermobility. In the group "possible hypermobile", 31 children (35%, 95%CI = 25 to 45%) were found to be hypermobile whilst in the group referred from a rheumatologist/paediatrician ("known hypermobile"), 11 children (52%, 95%CI = 31 to 74%) were Beighton positive.
The percentage of children at each level of the Beighton score was used in order to see if the distribution of the score across a group could differentiate between the three groups of children. Figure 1 suggested that the Beighton score was not able to clearly differentiate between the three groups of children with the "normals" and "possible hypermobiles" having a similar distribution. At the higher scores, the percentage of children increased in the "known hypermobile" group and decreased in the other two groups showing some distinction between the groups.
A Kruskal-Wallis test was used to test the null hypothesis of no significant difference being detectable between the three groups. The null hypothesis was rejected: the probability value obtained suggested that the difference between the groups was just on the level of significance ($\chi^2 = 5.87$, df = 2, p = 0.053).

Considering if the Lower Limb Assessment Score is a reasonable measure of hypermobility in children
The Lower Limb Assessment Score was investigated with regard to differentiating between the three groups of children. Observation of figure 2 suggested that the LLAS was able to distinguish more clearly between the groups. A Kruskal-Wallis test was used to test the null hypothesis of no significant difference occurring between the three groups of children. The null hypothesis was rejected: a significant difference was seen between the groups ($\chi^2 = 21.52$, df = 2, p < 0.001).

Defining a cut-off score to represent hypermobility with the LLAS
In order to consider the number of children that would be diagnosed with hypermobility using the Lower Limb Assessment Score, a threshold to define hypermobility had to be set. All chil-

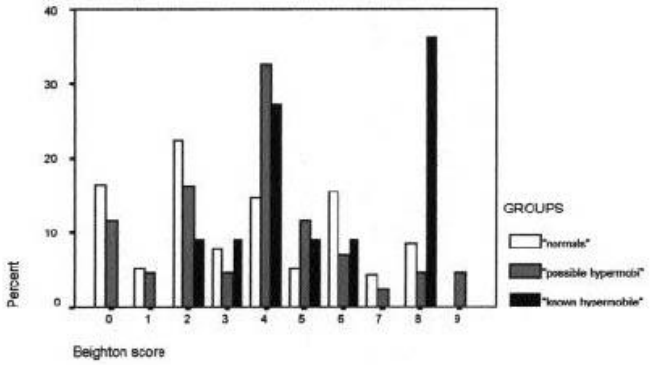

Fig. 1. Bar chart showing Beighton scores in three groups of children.

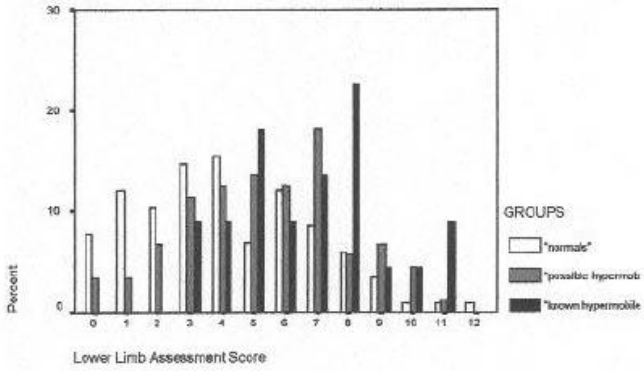

Fig. 2. Bar chart showing the Lower Limb Assessment Score for the three groups.

dren were assessed for the clinical opinion of the examiner as to whether they were hypermobile, borderline hypermobility or normal. The Lower Limb Assessment Score within each group was compared.

Figure 3 expresses these mean values graphically, with the thick bar representing the median value and the box defining the 25th and 75th percentile values. The bar ends represent the smallest and largest values that were not outliers.

A cut-off score of 8/12 appeared to give a score than would differentiate between hypermobile children and normal children, with minimal overlap with the borderline cases. Sensitivity analysis was undertaken to determine the numbers of true positive and false positive cases for each level of the score. The sensitivity and specificity for each level of the Lower Limb Assessment Score was tested using ROC analysis. For this analysis, the clinical opinion was used as the gold-standard for comparison. Children who were borderline cases were reclassified as normal for this analysis based upon the decision that if the clinician was undecided as to the diagnosis, the child was probably not hypermobile. Figure 4 shows the ROC curve and Table I gives the specificity and sensitivity levels.

The ROC curve (Fig. 4) and Table I shows how at the cut-off threshold of LLAS = 7, the sensitivity is good at 0.943 (94%) and the false positive rate is 0.075 (7.5%). At the threshold of LLAS = 8, the sensitivity has decreased to 0.704 (70.4%) but the false positive rate is extremely good at 0.004 (0.4%). The ROC curve shows the trade-off between sensitivity and specificity with the curve closest to the left-hand and top border being the most accurate. The point on the ROC curve that is closest to both axes expresses the most useful score in terms of sensitivity and specificity. On this curve, this point is the score of 7/12 for the LLAS.

The positive predictive values and negative predictive values were then considered in each group of children when a cut-off point of 7/12 was used to denote hypermobility. The positive predictive value tests the probability that the subject has hypermobility, when restricted to those subjects who do have hypermobility. Table II shows the values for each group.

With the cut-off score being identified, the percentage of children in each group found to be hypermobile could be identified. The percentages of children in each group when a cut-off point of 7/12 was applied were: "normals" 21% (95%CI=11 to25), "possible hypermobiles" 36% (95%CI= 26to 46) and "known hypermobile" 52% (95% CI=31to74). The percentages of children in each group when a cut-off point of 8/12 was applied were: "normals" 16% (95%CI=10to23), "possible hypermobiles" 38% (95%CI= 27to 48) and "known hypermobile" 41% (95% CI=21to64).

Comparing the Beighton score and the Lower Limb Assessment Score - can the Beighton score diagnose lower limb hypermobility? Does the LLAS fail to diagnose children with generalised hypermobility ?

The level of agreement between the two scores was sought using the Beighton score threshold for hypermobility as 5/9 and the LLAS threshold as 7/12. In the school children ("normals"), there was agreement between scoring systems in 69% of cases. In the remaining cases were there was disagreement, in 31 cases (26.7%), the Beighton score suggested the child was hypermobile

but the LLAS suggested that there was no hypermobility present in the lower limbs. In 5 cases (4.3%), the LLAS identified hypermobility but this was not accompanied by a Beighton score of greater than 5/9.

In the group "known hypermobile", there was agreement between the LLAS and the Beighton score in 80% of cases. In 2 children (10%) the Beighton score indicated hypermobility was present when the LLAS found no lower limb hypermobility. In 2 children (10%) the LLAS identified lower limb hypermobility but the Beighton score did not indicate generalised hypermobility.

The association between LLAS and the Beighton score was tested with Pearson correlation. A weak relationship was seen between the scores of the school children (r = 0.43) but a stronger relationship was seen for the "possible hypermobile" (r=0.79) and the "known hypermobile" groups (r = 0.79).

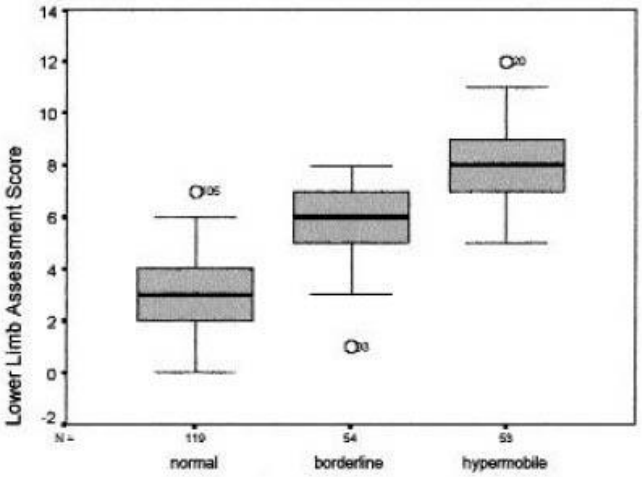

Fig. 3. Differences between the assessment scores for each clinical diagnosis (left side).

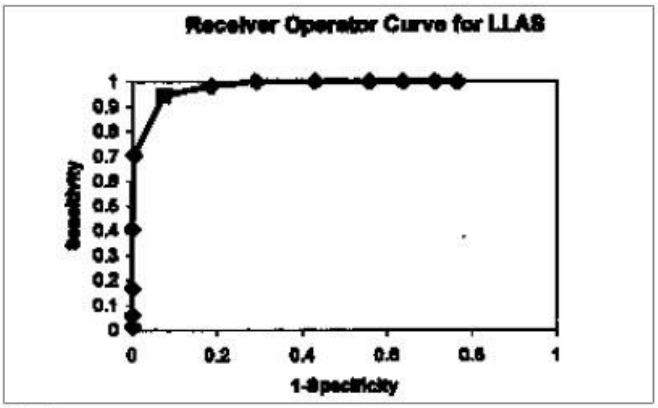

Fig. 4. ROC curve for the Lower Limb Assessment Score.

Discussion

The study found some differences between the scores that might be expected given that the scoring systems were measuring different aspects of hypermobility, but also placed some doubt on the use of the Beighton score in population studies. A difference in the levels of hypermobility was expected in the three populations. The "known hypermobiles" were expected to have the highest prevalence of hypermobility given that a diagnosis had been made prior to inclusion into the study, by a paediatrician or rheumatologist. The "possible hypermobiles" were expected to show a higher prevalence of hypermobility than the "normals" due to the numbers of cases of hypermobility seen in the clinics with foot or gait problems. The LLAS was clearly able to differentiate between these populations but the Beighton score could not differentiate between the groups so well. This may have been for two reasons. Firstly, two of the groups were seen in a foot and gait clinic so had problems presenting in the lower limbs – by measuring lower limb joints, the range of lower limb joint flexibility was going to be shown using the LLAS. By only measuring knee hypermobility, it was perhaps unfair to expect the Beighton score to distinguish between the particular groups. Secondly, the school children that were measured came from a predominantly Asian background whilst the children presenting to clinic were predominantly Caucasian. Asians are known to be more hypermobile than Caucasians (7) and thus the increased prevalence of hypermobility in these school children reduced the distinction between the "normals" and the "possible hypermobiles", with only the more extreme cases in the "known hypermobile" group remaining distinct.

The Beighton score appeared to overdiagnose hypermobility in the "normal" population in this study, demonstrated by the high prevalence of hypermobility identified (34%) when other studies have suggested that a figure of 15% to be more acceptable (8). Using a higher threshold or identifying an appropriate threshold through initial studies might reduce the prevalence seen. This was seen with the LLAS where

Table I. Sensitivity (true positive) and specificity (false positive) for diagnosing hypermobility with the LLAS.

Lower Limb Assessment Score		
Cut-off point	Sensitivity	1-specificity
0	1.000	0.765
1	1.000	0.712
2	1.000	0.637
3	1.000	0.558
4	1.000	0.429
5	1.000	0.292
6	0.981	0.186
7	0.943	0.075
8	0.704	0.004
9	0.407	0.000
10	0.169	0.000
11	0.058	0.000
12	0.014	0.000

Table II. Positive predictive values (PPV) and negative predictive values (NPV) for the three groups of children, taken at hypermobility = LLAS of 7/12 or greater.

	PPV	NPV
"Normals"	58.3%	89.1%
"Possible hypermobile"	84.4%	91.5%
"Known hypermobile"	83.3%	83.3%

sensitivity analysis allowed an appropriate threshold to be identified and the prevalence in the "normal" population was at a more acceptable level (21% using a threshold of 7/12 or 16% using a threshold of 8/12). Also suggestive that the Beighton score was over-diagnosing hypermobility at the 5/9 threshold was the level of disagreement between the Beighton and the LLAS in the "normal" population.

It was found that 26.7% of the school children ("normals") were positive for the Beighton score but negative for the LLAS. This would suggest that these children had upper limb/generalised hypermobility but no lower limb hypermobility. In the "known hypermobile" group there was 80% agreement between the scores so that most children with upper limb hypermobility also had lower limb hypermobility. It may be that the school children demonstrate a sub-population who have only upper limb hypermobility but it is more likely that the five tests used in the Beighton score were not sensitive enough to identify hypermobility, being easy to perform by many children. By only including one joint in the lower limb (knee hyperextension) the Beighton score also appeared to miss the diagnosis of hypermobility a small number of children (4.3%) who had lower limb hypermobility.

In life, flexibility is not an "all or none" state but there are degrees of flexibility and it is not possible to define one point when a person becomes hypermobile. However, in population or research studies it is useful to have a threshold for definition. If the LLAS was being used in a research project where and it was essential to exclude all normal children, then ROC analysis should be applied to identify an appropriate threshold for that population initially. Sensitivity and specificity can vary when populations are dramatically different and so it may be appropriate to repeat the analysis for older age groups or different ethnic groups.

Using the cut-off point of 7/12 for the LLAS in this study appeared to give more realistic values for the prevalence of hypermobility in the "normal" group (school children) of 21% but the prevalence of hypermobility in the "known hypermobile" group was low at 52% of the group compared with the Beighton score that found 60% of the group as hypermobile. For both these scoring systems, the prevalence of hypermobility in the "known hypermobile" group would be expected to be higher. It was unclear in the children being referred, how the diagnosis of hypermobility was made. It may have been based on the Beighton score and thus given the findings in the school group where the Beighton score was found to over-diagnose hypermobility, the LLAS would be expected to be lower. If this was the situation, the prevalence with the Beighton score should have been closer to 100%. The lower figure may reflect examiner bias where more strict criteria were applied when measuring the score for the study compared with a clinical situation. Alternatively, the referring practitioner may only have found hypermobility at a particular joint – for example, the symptomatic joint – and given the diagnosis based upon that joint and not applied any scoring system during their assessment.

In conclusion, this study introduced a new scoring system for the diagnosis of hypermobility in the lower limb joints. The scoring system was shown to have benefits over the traditionally used Beighton score and although best used as a sliding scale of flexibility, a cut-off point to diagnose hypermobility in the lower limb was set. The Lower Limb Assessment Score would be useful for prospective studies, when excluding/including hypermobile participants. It would be a useful clinical tool for health professionals to aid in the diagnosis of lower limb conditions that may be related to joint hypermobility.

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APPENDIX 1

Lower Limb Assessment Score	LEFT		RIGHT	
HIP FLEXION – The patient lies supine; the examiner flexes one hip fully; the other leg must stay fully extended on the couch. Does the mid-anterior area of the thigh drop easily onto the stomach/chest with a loose feel to the movement, using a minimum to moderate application of force?	YES	NO	YES	NO
HIP ABDUCTION – The patient lies supine, with hip and knees flexed; the knees are dropped outwards and down to the couch, the soles of the feet remain together. With the examiner's hand against the lateral femoral condyle, can the knees come down to the couch sufficiently to let the back of the examiners hand touch the couch? - minimal application of force required.	YES	NO	YES	NO
KNEE HYPEREXTENSION – The patient lies supine; the knees are relaxed and straight; With minimal force, keeping the femoral condyles on the couch, can the heel be lifted at least 3cm off the couch (greater than 2 finger widths)?	YES	NO	YES	NO
KNEE ANTERIOR DRAW TEST – The patient is supine; the hips and knees (90°) are flexed; the examiner gently sits of the foot to stabilise it; moderate pressure is placed against the femoral condyles as the tibia is pulled forwards. Is there a definite, obvious forward movement of the tibia against the femur? Palpable "clunking" of the joint surfaces moving against each is indicative of a positive draw sign.	YES	NO	YES	NO
KNEE ROTATION – The patient lies supine; the examiner flexes the hip and knee to 90° and palpates the tibial tubercle; holding the malleoli and ankle firmly, the tibia is rotated medially and laterally on the femur. Normal movement is 1cm medially and laterally. Does the tubercle move easily beyond 1cm in any direction or greater than 2cm overall? With increased internal movement the head of the fibula/lateral condyle of the tibia may also be seen to move.	YES	NO	YES	NO
ANKLE JOINTDORSIFLEXION – The patient lies supine; the knee is flexed to 45°; with moderate to strong force the ankle is dorsiflexed. Does the ankle flex more than 15 degrees? Along with the increased movement there may be bulging of the skin and subcutaneous fat anterior to the ankle.	YES	NO	YES	NO
ANKLE ANTERIOR DRAW TEST – The patient lies supine; the knee is flexed to 45°; the examiner grasps the heel along the plantar and posterior surfaces with one hand and applied a stabilising force against the anterior of the tibia with the other hand. Using a strong anterior force, can the calcaneum and talus be brought forwards on the tibia? Any forward movement felt is a positive result.	YES	NO	YES	NO
SUBTALAR JOINTINVERSION – The patient is supine with their feet over the end of the couch; the examiner holds the posterior surface of the heel and moves the heel into inversion without moving the leg. Is excessive inversion of the subtalar joint seen using minimal force? The sole of the foot or visualisation of the neck of the talus should show movement of 45° inwards, the lateral head of the talus will be very prominent.	YES	NO	YES	NO
MIDTARSAL JOINT INVERSION – The patient is supine with their feet over the end of the couch; the midtarsal joint is isolated from the subtalar joint; the forefoot is grasped from lateral to medial along the metatarsals; only minimal - moderate force is applied to invert the midtarsal joint. Does the midtarsal joint invert beyond 45° so that the plantar surface of the metatarsal heads can be brought inwards by 45 degrees?	YES	NO	YES	NO
MIDTARSAL JOINT AB/ADDUCTION AND DORSI/PLANTARFLEXION – The patient is supine with their feet over the end of the couch; the examiner grasps and stabilises the rearfoot; the forefoot is moved in the direction of ab/adduction and dorsi/plantarflexion. Normal movement should be 1cm in each direction. With minimal force, does the forefoot move easily, almost "wobbling", in an increased amount? Excessive movement in either of the two planes is a positive result.	YES	NO	YES	NO
METATARSOPHALANGEAL MOVEMENT – The patient is supine with their feet over the end of the couch; the hallux is dorsiflexed using minimal – moderate force. Does the hallux dorsiflex easily beyond 90° relative to the metatarsal?	YES	NO	YES	NO

APPENDIX 1

LowerLimb Assessment Score	LEFT		RIGHT	
EXCESSIVE SUBTALAR JOINTPRONATION – The patient is to march on the spot and stop on command; the patient is asked to invert their foot and hold the position close to subtalar joint neutral; the patient is then asked to relax their foot; the movement is observed. Does the arch lower and flatten fully, excessively and easily, with the talus bulging medially ? The pronation noted should be at the end of range of the subtalar joint motion so that no further pronation is possible	YES	NO	YES	NO
To score, each limb is calculated separately giving a left score and right score. Each YES is given one mark. Atotal of score of 12 marks is available.	TOTAL:			

Beighton Score [taken from Beighton *et al.* (2) and Hudson *et al.* (10)].				
PASSIVE DORSIFLEXION OF THE 5TH FINGER M.C.P JOINT – With the palm placed on a flat surface, the 5th finger is raised so that the m.c.p joint is dorsiflexed to resistance using miminal force. An angle of 90 degrees or greater is a positive result.	YES	NO	YES	NO
PASSIVE APPOSITION OF THE THUMB TO THE FLEXOR ASPECT OF THE FOREARM – With the wrist flexed, the thumb is passively moved towards the forearm. The thumb contacting the forearm using minimal force is a positive result.	YES	NO	YES	NO
HYPEREXTENSION OF THE ELBOW – With the subject sitting or standing and the upper arm supported, the forearm is aligned with the upper arm with the elbow in a neutral position. The forearm is then allowed to drop and gentle pressure placed on the forearm to hyperextend the elbow. Extension greater than 10 degrees is a positive result.	YES	NO	YES	NO
HYPEREXTENSION OF THE KNEE – The subject lies supine and the knees relaxed and straight. With minimal force, keeping the femoral condyles on the couch, can the heel be lifted at least 3cm off the couch – 10 degrees hyperextension?	YES	NO	YES	NO
FORWARD FLEXION OF THE TRUNK – With the subject standing, keeping the knees straight, the subject is asked to bend forwards and touch the floor. Placing the palms of the hands on the floor is a positive result.	YES	NO	YES	NO
	One point is given for each "yes", to give a total score out of 9.			

Appendix 27: Foot Posture Index

THE FOOT POSTURE INDEX©
FPI-6

Reference Sheet

The patient should stand in their relaxed stance position with double limb support. The patient should be instructed to stand still, with their arms by the side and looking straight ahead. It may be helpful to ask the patient to take several steps, marching on the spot, prior to settling into a comfortable stance position. During the assessment, it is important to ensure that the patient does not swivel to try to see what is happening for themself, as this will significantly affect the foot posture. The patient will need to stand still for approximately two minutes in total in order for the assessment to be conducted. The assessor needs to be able to move around the patient during the assessment and to have uninterrupted access to the posterior aspect of the leg and foot.

If an observation cannot be made (e.g. because of soft tissue swelling) simply miss it out and indicate on the datasheet that the item was not scored.

If there is genuine doubt about how high or low to score an item always use the more conservative score.

Rearfoot Score	**-2**	**-1**	**0**	**1**	**2**
Talar head palpation	Talar head palpable on lateral side/but not on medial side	Talar head palpable on lateral side/slightly palpable on medial side	Talar head equally palpable on lateral and medial side	Talar head slightly palpable on lateral side/ palpable on medial side	Talar head not palpable on lateral side/ but palpable on medial side
Curves above and below the malleoli	Curve below the malleolus either straight or convex	Curve below the malleolus concave, but flatter/ more shallow than the curve above the malleolus	Both infra and supra malleolar curves roughly equal	Curve below malleolus more concave than curve above malleolus	Curve below malleolus markedly more concave than curve above malleolus
Calcaneal inversion/eversion	More than an estimated 5° inverted (varus)	Between vertical and an estimated 5° inverted (varus)	Vertical	Between vertical and an estimated 5° everted (valgus)	More than an estimated 5° everted (valgus)
Forefoot Score	**-2**	**-1**	**0**	**1**	**2**
Talo-navicular congruence	Area of TNJ markedly concave	Area of TNJ slightly, but definitely concave	Area of TNJ flat	Area of TNJ bulging slightly	Area of TNJ bulging markedly
Medial arch height	Arch high and acutely angled towards the posterior end of the medial arch	Arch moderately high and slightly acute posteriorly	Arch height normal and concentrically curved	Arch lowered with some flattening in the central portion	Arch very low with severe flattening in the central portion – arch making ground contact
Forefoot abd/adduction	No lateral toes visible. Medial toes clearly visible	Medial toes clearly more visible than lateral	Medial and lateral toes equally visible	Lateral toes clearly more visible than medial	No medial toes visible. Lateral toes clearly visible

For further information, manuals and extra datasheets see: www.leeds.ac.uk/medicine/FASTER/FPI/

Foot Posture Index Datasheet

Patient name	***ID number***

	FACTOR	*PLANE*	SCORE 1 Date_____ Comment_____		SCORE 2 Date_____ Comment_____		SCORE 3 Date_____ Comment_____	
			Left *-2 to +2*	*Right* *-2 to +2*	*Left* *-2 to +2*	*Right* *-2 to +2*	*Left* *-2 to +2*	*Right* *-2 to +2*
Rearfoot	Talar head palpation	*Transverse*						
	Curves above and below the lateral malleolus	*Frontal/ transverse*						
	Inversion/eversion of the calcaneus	*Frontal*						
Forefoot	Prominence in the region of the TNJ	*Transverse*						
	Congruence of the medial longitudinal arch	*Sagittal*						
	Abd/adduction forefoot on rearfoot	*Transverse*						
	TOTAL							

Reference values
Normal = 0 to +5
Pronated = +6 to +9, Highly pronated 10+
Supinated = -1 to –4, Highly supinated –5 to -12

Foot Posture Index Datasheet

Patient name	***ID number***

	FACTOR	*PLANE*	SCORE 1 Date_____ Comment_____		SCORE 2 Date_____ Comment_____		SCORE 3 Date_____ Comment_____	
			Left *-2 to +2*	*Right* *-2 to +2*	*Left* *-2 to +2*	*Right* *-2 to +2*	*Left* *-2 to +2*	*Right* *-2 to +2*
Rearfoot	Talar head palpation	*Transverse*						
	Curves above and below the lateral malleolus	*Frontal/ transverse*						
	Inversion/eversion of the calcaneus	*Frontal*						
Forefoot	Prominence in the region of the TNJ	*Transverse*						
	Congruence of the medial longitudinal arch	*Sagittal*						
	Abd/adduction forefoot on rearfoot	*Transverse*						
	TOTAL							

Reference values
Normal = 0 to +5
Pronated = +6 to +9, Highly pronated 10+
Supinated = -1 to –4, Highly supinated –5 to -12

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Appendix 28: Extra Results Chapter 4

PedsQL™ 4.0 Generic Core Quality of Life

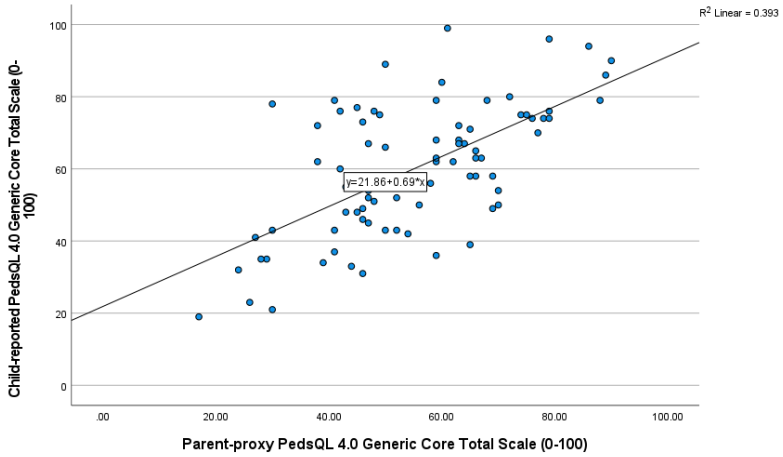


Figure. Parent and child-reported total fatigue as measured by PedsQL 4.0 Generic Core total scale (0-100) where higher scores indicate better quality of life.

Pain

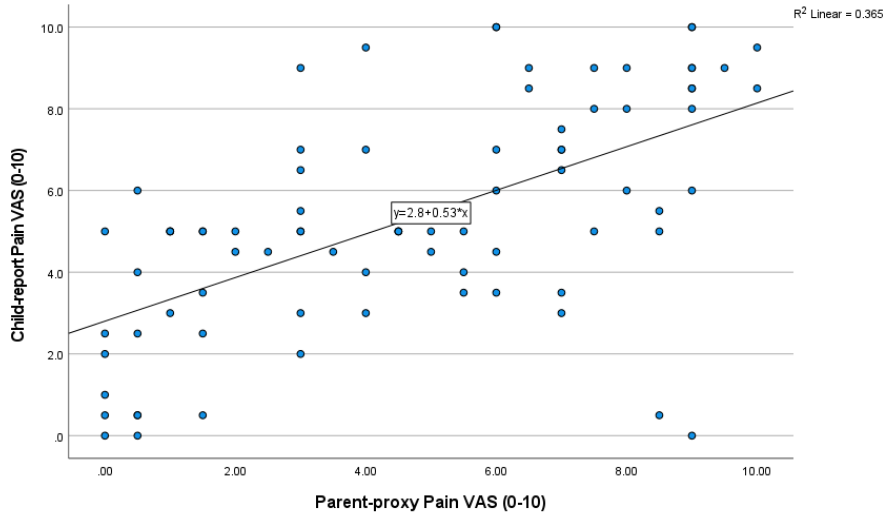


Figure. Child-reported and parent proxy report Pain VAS. VAS Visual Analogue Scale 0-10 where higher scores indicate worse pain.

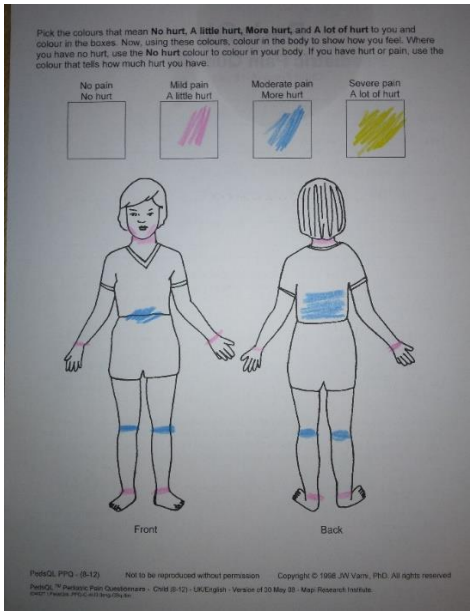


Figure. Children aged 5-7 years and 8-12 years coloured in a body outline with the selected colour/intensity match.

PedsQL Multidimensional fatigue

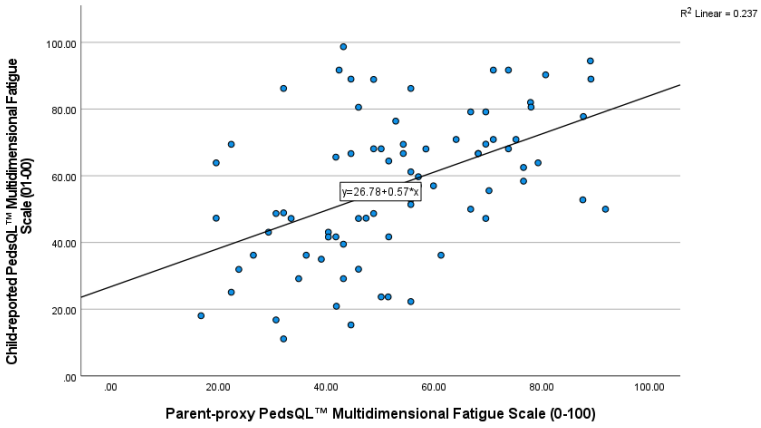


Figure. Child-reported and parent proxy report PedsQL™ Multidimensional Fatigue Scale. PedsQL™ Multidimensional Fatigue Scale score 0-100 where higher number indicates less fatigue.

Cluster analysis

Descriptive Statistics

	Mean	Std. Deviation	N
total_fatig_child	57.1644	21.90187	80
total_qol_child	60.03	18.228	80
pain_worst_child	5.325	2.7868	80
PA_score	2.3888	.71747	80

Figure. Mean and standard deviation for the four questionnaire measures n=80

Descriptive Statistics

	Mean	Std. Deviation	N
total_fatig_child	57.1644	21.90187	80
total_qol_child	60.03	18.228	80
pain_worst_child	5.325	2.7868	80
PA_score	2.3888	.71747	80
funct_6MWD_m	518.56	91.048	80
YBT_both_cm	100.3159	12.96782	73
strength_avg_kg	10.3792	6.34548	80
msl_endurance_s	66.29	40.618	80
length_popangle	145.88	13.934	80
motskills_std_score	42.30	10.176	79
beightonscore	6.01	1.775	80
LLAS	6.71	3.098	80
foot_post_index	6.981	2.6784	80

Figure. Mean and standard deviation of the nine objective measures n=80

Statistics

		funct_6MWD_m	YBT_both_cm	LLAS	beightonscore	foot_post_index	msl_endurance_s	length_popangle	strength_avg_kg	motskills_std_score
N	Valid	80	73	80	80	80	80	80	80	79
	Missing	0	7	0	0	0	0	0	0	1
Mean		518.56	100.3159	6.71	6.01	6.981	66.29	145.88	10.3792	42.30
Median		517.50	99.7000	7.00	6.00	7.250	52.00	140.00	9.9167	42.00
Std. Deviation		91.048	12.96782	3.098	1.775	2.6784	40.618	13.934	6.34548	10.176
Percentiles	25	465.50	92.1500	5.00	6.00	5.000	31.00	135.00	5.4583	36.00
	50	517.50	99.7000	7.00	6.00	7.250	52.00	140.00	9.9167	42.00
	75	570.00	107.4600	9.00	7.00	9.000	120.00	158.75	14.8646	47.00

Figure. Descriptive statistics of the nine objective measures n = 80

Correlations (bivariate) between all features

Correlations

		total_fatig_child	total_qol_child	pain_worst_child	PA_score
total_fatig_child	Pearson Correlation	1	.819**	-.540**	.521**
	Sig. (2-tailed)		.000	.000	.000
	N	80	80	80	80
total_qol_child	Pearson Correlation	.819**	1	-.571**	.470**
	Sig. (2-tailed)	.000		.000	.000
	N	80	80	80	80
pain_worst_child	Pearson Correlation	-.540**	-.571**	1	-.180
	Sig. (2-tailed)	.000	.000		.110
	N	80	80	80	80
PA_score	Pearson Correlation	.521**	.470**	-.180	1
	Sig. (2-tailed)	.000	.000	.110	
	N	80	80	80	80

**. Correlation is significant at the 0.01 level (2-tailed).

Figure. Correlation (bivariate) in questionnaire features

Correlations

		funct_6MWD_m	YBT_both_cm	strength_avg_kg	length_popangle	motskills_std_score	beightonscore	LLAS	foot_post_index
funct_6MWD_m	Pearson Correlation	1	.164	.010	-.162	.280*	-.064	.076	-.070
	Sig. (2-tailed)		.165	.931	.152	.013	.575	.502	.536
	N	80	73	80	80	79	80	80	80
YBT_both_cm	Pearson Correlation	.164	1	.158	.292*	.460**	.138	-.012	-.268*
	Sig. (2-tailed)	.165		.183	.012	.000	.243	.919	.022
	N	73	73	73	73	73	73	73	73
strength_avg_kg	Pearson Correlation	.010	.158	1	-.014	.197	-.284*	-.194	-.239*
	Sig. (2-tailed)	.931	.183		.901	.082	.011	.085	.033
	N	80	73	80	80	79	80	80	80
length_popangle	Pearson Correlation	-.162	.292*	-.014	1	.275*	.337**	.082	-.256*
	Sig. (2-tailed)	.152	.012	.901		.014	.002	.469	.022
	N	80	73	80	80	79	80	80	80
motskills_std_score	Pearson Correlation	.280*	.460**	.197	.275*	1	.155	.200	-.064
	Sig. (2-tailed)	.013	.000	.082	.014		.172	.078	.575
	N	79	73	79	79	79	79	79	79
beightonscore	Pearson Correlation	-.064	.138	-.284*	.337**	.155	1	.394**	.105
	Sig. (2-tailed)	.575	.243	.011	.002	.172		.000	.353
	N	80	73	80	80	79	80	80	80
LLAS	Pearson Correlation	.076	-.012	-.194	.082	.200	.394**	1	.408**
	Sig. (2-tailed)	.502	.919	.085	.469	.078	.000		.000
	N	80	73	80	80	79	80	80	80
foot_post_index	Pearson Correlation	-.070	-.268*	-.239*	-.256*	-.064	.105	.408**	1
	Sig. (2-tailed)	.536	.022	.033	.022	.575	.353	.000	
	N	80	73	80	80	79	80	80	80

*. Correlation is significant at the 0.05 level (2-tailed).

**. Correlation is significant at the 0.01 level (2-tailed).

Figure. Correlation (bivariate) in objective features

Correlations

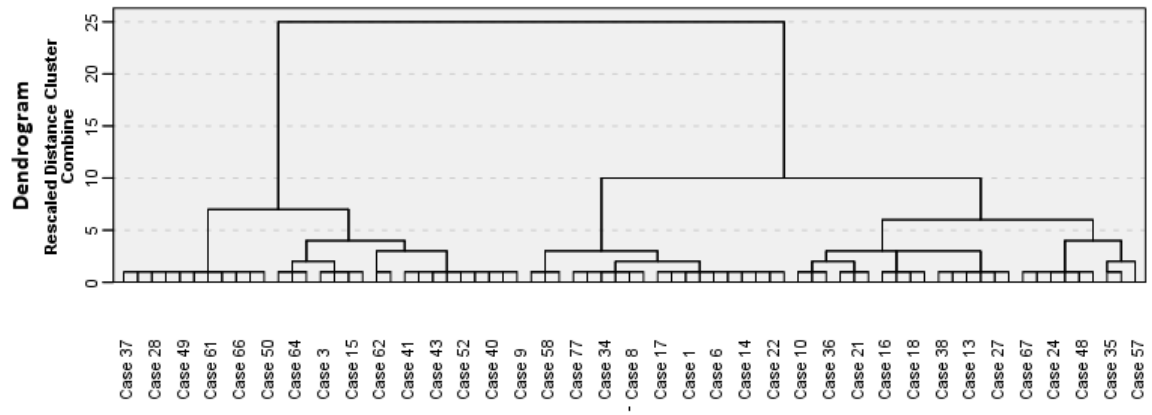
		total_fatig_child	total_qol_child	pain_worst_child	PA_score	funct_6MWD_m	YBT_both_cm	strength_avg_kg	msl_endurance_s	musicle length	motskills_std_score	beightonscore	LLAS	foot_post_index
total_fatig_child	Pearson Correlation	1	.819**	-.540**	.521**	.384**	.243*	-.038	.342**	-.051	.327**	-.055	.076	.162
	Sig. (2-tailed)		.000	.000	.000	.000	.038	.738	.002	.652	.003	.626	.501	.152
	N	80	80	80	80	80	73	80	80	80	79	80	80	80
total_qol_child	Pearson Correlation	.819**	1	-.571**	.470**	.399**	.250*	.014	.290**	-.044	.345**	-.032	.143	.160
	Sig. (2-tailed)	.000		.000	.000	.000	.033	.902	.009	.699	.002	.778	.205	.156
	N	80	80	80	80	80	73	80	80	80	79	80	80	80
pain_worst_child	Pearson Correlation	-.540**	-.571**	1	-.180	-.229*	-.284*	.052	-.158	.024	-.322**	-.110	-.163	-.201
	Sig. (2-tailed)	.000	.000		.110	.041	.015	.650	.162	.836	.004	.333	.149	.074
	N	80	80	80	80	80	73	80	80	80	79	80	80	80
PA_score	Pearson Correlation	.521**	.470**	-.180	1	.529**	.402**	.066	.371**	.001	.275*	.050	.047	-.134
	Sig. (2-tailed)	.000	.000	.110		.000	.000	.560	.001	.995	.014	.662	.681	.235
	N	80	80	80	80	80	73	80	80	80	79	80	80	80
funct_6MWD_m	Pearson Correlation	.384**	.399**	-.229*	.529**	1	.164	.010	.360**	-.162	.280*	-.064	.076	-.070
	Sig. (2-tailed)	.000	.000	.041	.000		.165	.931	.001	.152	.013	.575	.502	.536
	N	80	80	80	80	80	73	80	80	80	79	80	80	80
YBT_both_cm	Pearson Correlation	.243*	.250*	-.284*	.402**	.164	1	.158	.357**	.292*	.460**	.138	-.012	-.268*
	Sig. (2-tailed)	.038	.033	.015	.000	.165		.183	.002	.012	.000	.243	.919	.022
	N	73	73	73	73	73	73	73	73	73	73	73	73	73
strength_avg_kg	Pearson Correlation	-.038	.014	.052	.066	.010	.158	1	.401**	-.014	.197	-.284*	-.194	-.239*
	Sig. (2-tailed)	.738	.902	.650	.560	.931	.183		.000	.901	.082	.011	.085	.033
	N	80	80	80	80	80	73	80	80	80	79	80	80	80
msl_endurance_s	Pearson Correlation	.342**	.290**	-.158	.371**	.360**	.357**	.401**	1	.134	.363**	-.162	.047	-.132
	Sig. (2-tailed)	.002	.009	.162	.001	.001	.002	.000		.236	.001	.152	.679	.245
	N	80	80	80	80	80	73	80	80	80	79	80	80	80
length_popangle	Pearson Correlation	-.051	-.044	.024	.001	-.162	.292*	-.014	.134	1	.275*	.337**	.082	-.256*
	Sig. (2-tailed)	.652	.699	.836	.995	.152	.012	.901	.236		.014	.002	.469	.022
	N	80	80	80	80	80	73	80	80	80	79	80	80	80
motskills_std_score	Pearson Correlation	.327**	.345**	-.322**	.275*	.280*	.460**	.197	.363**	.275*	1	.155	.200	-.064
	Sig. (2-tailed)	.003	.002	.004	.014	.013	.000	.082	.001	.014		.172	.078	.575
	N	79	79	79	79	79	73	79	79	79	79	79	79	79
beightonscore	Pearson Correlation	-.055	-.032	-.110	.050	-.064	.138	-.284*	-.162	.337**	.155	1	.394**	.105
	Sig. (2-tailed)	.626	.778	.333	.662	.575	.243	.011	.152	.002	.172		.000	.353
	N	80	80	80	80	80	73	80	80	80	79	80	80	80
LLAS	Pearson Correlation	.076	.143	-.163	.047	.076	-.012	-.194	.047	.082	.200	.394**	1	.408**
	Sig. (2-tailed)	.501	.205	.149	.681	.502	.919	.085	.679	.469	.078	.000		.000
	N	80	80	80	80	80	73	80	80	80	79	80	80	80
foot_post_index	Pearson Correlation	.162	.160	-.201	-.134	-.070	-.268*	-.239*	-.132	-.256*	-.064	.105	.408**	1
	Sig. (2-tailed)	.152	.156	.074	.235	.536	.022	.033	.245	.022	.575	.353	.000	
	N	80	80	80	80	80	73	80	80	80	79	80	80	80

**. Correlation is significant at the 0.01 level (2-tailed).

*. Correlation is significant at the 0.05 level (2-tailed).

Figure. Correlation (bivariate) in all questionnaire and objective features

Dendrogram



Participants

Figure. Dendrogram of five-cluster feature model. The x-axis (leaves) indicate individual patients, segments on the height (y-axis) indicate distances where clusters are combined. This is dendrogram is combined at 5 on the y-axis which indicates the five-cluster solution

Subgroup comparison

Descriptives

		N	Mean	Std. Deviation	Std. Error	95% Confidence Interval for Mean		Minimum	Maximum
						Lower Bound	Upper Bound		
motskills_std_score	Medium Intervention Subgroup	42	39.71	6.656	1.027	37.64	41.79	23	52
	Low Intervention Subgroup	16	57.75	6.330	1.582	54.38	61.12	50	71
	High Intervention Subgroup	21	35.71	5.693	1.242	33.12	38.31	20	43
	Total	79	42.30	10.176	1.145	40.02	44.58	20	71
total_fatig_child	Medium Intervention Subgroup	42	63.4209	18.12916	2.79739	57.7715	69.0703	22.30	98.70
	Low Intervention Subgroup	16	68.1500	18.52271	4.63068	58.2799	78.0201	39.50	94.44
	High Intervention Subgroup	21	35.1677	15.79702	3.44719	27.9770	42.3584	11.11	61.20
	Total	79	56.8684	21.88009	2.46170	51.9675	61.7692	11.11	98.70
pain_worst_child	Medium Intervention Subgroup	42	3.964	1.9518	.3012	3.356	4.573	.0	7.0
	Low Intervention Subgroup	16	4.656	2.1735	.5434	3.498	5.814	.5	8.0
	High Intervention Subgroup	21	8.810	.9549	.2084	8.375	9.244	6.5	10.0
	Total	79	5.392	2.7382	.3081	4.779	6.006	.0	10.0

Figure. Descriptive data three cluster features motor skills proficiency, total fatigue, and worst pain

One-way analysis of variance ANOVA

ANOVA

		Sum of Squares	df	Mean Square	F	Sig.
motskills_std_score	Between Groups	5010.852	2	2505.426	62.107	.000
	Within Groups	3065.857	76	40.340		
	Total	8076.709	78			
total_fatig_child	Between Groups	13728.978	2	6864.489	22.094	.000
	Within Groups	23612.610	76	310.692		
	Total	37341.588	78			
pain_worst_child	Between Groups	339.542	2	169.771	52.600	.000
	Within Groups	245.294	76	3.228		
	Total	584.835	78			

Figure. One-way analysis of variance (ANOVA) to compare three cluster features motor skills proficiency, total fatigue, and worst pain

Descriptives

		N	Mean	Std. Deviation	Std. Error	95% Confidence Interval for Mean		Minimum	Maximum
						Lower Bound	Upper Bound		
age	Medium Intervention Subgroup	42	11.7250	2.63501	.40659	10.9039	12.5461	6.08	16.83
	Low Intervention Subgroup	16	10.7192	3.59061	.89765	8.8059	12.6325	6.00	16.92
	High Intervention Subgroup	21	12.0432	3.24926	.70905	10.5641	13.5222	6.83	16.83
	Total	79	11.6059	3.00940	.33858	10.9318	12.2799	6.00	16.92
BMI_centile	Medium Intervention Subgroup	42	54.29	29.694	4.582	45.03	63.54	1	96
	Low Intervention Subgroup	16	55.38	30.135	7.534	39.32	71.43	4	96
	High Intervention Subgroup	21	74.29	22.963	5.011	63.83	84.74	20	99
	Total	79	59.82	29.158	3.281	53.29	66.35	1	99
time_diagnosis_months	Medium Intervention Subgroup	42	35.7857	24.25830	3.74314	28.2263	43.3451	3.00	108.00
	Low Intervention Subgroup	16	21.1875	13.74636	3.43659	13.8626	28.5124	3.00	48.00
	High Intervention Subgroup	21	32.3333	24.47720	5.34136	21.1914	43.4752	3.00	90.00
	Total	79	31.9114	23.04354	2.59260	26.7499	37.0729	3.00	108.00
beightonscore	Medium Intervention Subgroup	42	5.76	1.845	.285	5.19	6.34	0	8
	Low Intervention Subgroup	16	6.50	2.000	.500	5.43	7.57	3	9
	High Intervention Subgroup	21	6.19	1.436	.313	5.54	6.84	2	8
	Total	79	6.03	1.783	.201	5.63	6.42	0	9
LLAS	Medium Intervention Subgroup	42	6.43	3.085	.476	5.47	7.39	0	11
	Low Intervention Subgroup	16	7.94	2.323	.581	6.70	9.18	3	11
	High Intervention Subgroup	21	6.38	3.584	.782	4.75	8.01	0	10
	Total	79	6.72	3.117	.351	6.02	7.42	0	11
foot_post_index	Medium Intervention Subgroup	42	7.107	2.8253	.4359	6.227	7.988	-1.0	11.5
	Low Intervention Subgroup	16	6.875	2.7049	.6762	5.434	8.316	1.0	10.5
	High Intervention Subgroup	21	6.571	2.2377	.4883	5.553	7.590	3.5	11.0
	Total	79	6.918	2.6341	.2964	6.328	7.508	-1.0	11.5
length_popangle	Medium Intervention Subgroup	42	143.57	11.492	1.773	139.99	147.15	120	170
	Low Intervention Subgroup	16	153.13	17.783	4.446	143.65	162.60	125	180
	High Intervention Subgroup	21	145.71	13.900	3.033	139.39	152.04	125	170
	Total	79	146.08	13.906	1.565	142.96	149.19	120	180
motskills_centile	Medium Intervention Subgroup	42	19.64	13.875	2.141	15.32	23.97	1	58
	Low Intervention Subgroup	16	74.19	14.901	3.725	66.25	82.13	50	98
	High Intervention Subgroup	21	10.71	8.174	1.784	6.99	14.44	1	29
	Total	79	28.32	26.769	3.012	22.32	34.31	1	98
total_qol_child	Medium Intervention Subgroup	42	65.95	16.000	2.469	60.97	70.94	21	99
	Low Intervention Subgroup	16	67.75	14.866	3.717	59.83	75.67	36	96
	High Intervention Subgroup	21	43.10	13.601	2.968	36.90	49.29	19	78
	Total	79	60.24	18.242	2.052	56.15	64.33	19	99
funct_6MWD_m	Medium Intervention Subgroup	42	527.76	86.887	13.407	500.69	554.84	372	758
	Low Intervention Subgroup	16	550.50	76.602	19.151	509.68	591.32	440	682
	High Intervention Subgroup	21	474.81	99.201	21.647	429.65	519.97	210	597
	Total	79	518.29	91.598	10.306	497.77	538.81	210	758
YBT_both_cm	Medium Intervention Subgroup	39	99.2956	14.58998	2.33627	94.5661	104.0252	69.82	144.07
	Low Intervention Subgroup	16	106.7706	11.74729	2.93682	100.5109	113.0303	94.75	143.07
	High Intervention Subgroup	18	96.7889	7.73747	1.82374	92.9411	100.6366	84.37	109.12
	Total	73	100.3159	12.96782	1.51777	97.2903	103.3415	69.82	144.07
msl_endurance_s	Medium Intervention Subgroup	42	65.57	37.278	5.752	53.95	77.19	12	120
	Low Intervention Subgroup	16	85.00	40.211	10.053	63.57	106.43	19	120
	High Intervention Subgroup	21	54.00	44.826	9.782	33.60	74.40	12	120
	Total	79	66.43	40.858	4.597	57.28	75.58	12	120
PA_score	Medium Intervention Subgroup	42	2.3812	.68519	.10573	2.1677	2.5948	1.31	4.18
	Low Intervention Subgroup	16	2.6570	.81658	.20414	2.2218	3.0921	1.56	4.22
	High Intervention Subgroup	21	2.1574	.64643	.14106	1.8631	2.4516	1.22	3.91
	Total	79	2.3776	.71500	.08044	2.2174	2.5377	1.22	4.22

Figure. Descriptive data all variables by subgroup

ANOVA

		Sum of Squares	df	Mean Square	F	Sig.
age	Between Groups	17.192	2	8.596	.948	.392
	Within Groups	689.214	76	9.069		
	Total	706.406	78			
BMI_centile	Between Groups	5996.912	2	2998.456	3.778	.027
	Within Groups	60318.607	76	793.666		
	Total	66315.519	78			
time_diagnosis_months	Between Groups	2474.204	2	1237.102	2.414	.096
	Within Groups	38944.176	76	512.423		
	Total	41418.380	78			
beightonscore	Between Groups	7.092	2	3.546	1.119	.332
	Within Groups	240.857	76	3.169		
	Total	247.949	78			
LLAS	Between Groups	29.698	2	14.849	1.550	.219
	Within Groups	728.176	76	9.581		
	Total	757.873	78			
foot_post_index	Between Groups	4.054	2	2.027	.287	.751
	Within Groups	537.161	76	7.068		
	Total	541.215	78			
length_popangle	Between Groups	1061.223	2	530.611	2.876	.063
	Within Groups	14022.321	76	184.504		
	Total	15083.544	78			
motskills_centile	Between Groups	43332.723	2	21666.361	131.098	.000
	Within Groups	12560.366	76	165.268		
	Total	55893.089	78			
total_qol_child	Between Groups	8445.716	2	4222.858	18.328	.000
	Within Groups	17510.714	76	230.404		
	Total	25956.430	78			
funct_6MWD_m	Between Groups	60069.447	2	30034.723	3.840	.026
	Within Groups	594360.857	76	7820.538		
	Total	654430.304	78			
YBT_both_cm	Between Groups	931.128	2	465.564	2.916	.061
	Within Groups	11176.713	70	159.667		
	Total	12107.841	72			
msl_endurance_s	Between Groups	8793.081	2	4396.541	2.752	.070
	Within Groups	121416.286	76	1597.583		
	Total	130209.367	78			
PA_score	Between Groups	2.268	2	1.134	2.291	.108
	Within Groups	37.608	76	.495		
	Total	39.876	78			

Figure. One-way analysis of variance (ANOVA) to compare all variables

Tukey HSD Post Hoc Tests

Multiple Comparisons

Tukey HSD

Dependent Variable	(I) Ward Method	(J) Ward Method	Mean Difference (I-J)	Std. Error	Sig.	95% Confidence Interval	
						Lower Bound	Upper Bound
age	Medium Intervention Subgroup	Low Intervention Subgroup	1.00583	.88471	.495	-1.1090	3.1207
		High Intervention Subgroup	-.31817	.80483	.918	-2.2421	1.6058
	Low Intervention Subgroup	Medium Intervention Subgroup	-1.00583	.88471	.495	-3.1207	1.1090
		High Intervention Subgroup	-1.32401	.99931	.386	-3.7128	1.0648
	High Intervention Subgroup	Medium Intervention Subgroup	.31817	.80483	.918	-1.6058	2.2421
		Low Intervention Subgroup	1.32401	.99931	.386	-1.0648	3.7128
BMI_centile	Medium Intervention Subgroup	Low Intervention Subgroup	-1.089	8.277	.990	-20.87	18.70
		High Intervention Subgroup	-20.000*	7.529	.026	-38.00	-2.00
	Low Intervention Subgroup	Medium Intervention Subgroup	1.089	8.277	.990	-18.70	20.87
		High Intervention Subgroup	-18.911	9.349	.114	-41.26	3.44
	High Intervention Subgroup	Medium Intervention Subgroup	20.000*	7.529	.026	2.00	38.00
		Low Intervention Subgroup	18.911	9.349	.114	-3.44	41.26
time_diagnosis_months	Medium Intervention Subgroup	Low Intervention Subgroup	14.59821	6.65034	.078	-1.2993	30.4957
		High Intervention Subgroup	3.45238	6.04993	.836	-11.0099	17.9146
	Low Intervention Subgroup	Medium Intervention Subgroup	-14.59821	6.65034	.078	-30.4957	1.2993
		High Intervention Subgroup	-11.14583	7.51183	.304	-29.1027	6.8110
	High Intervention Subgroup	Medium Intervention Subgroup	-3.45238	6.04993	.836	-17.9146	11.0099
		Low Intervention Subgroup	11.14583	7.51183	.304	-6.8110	29.1027
beightonscore	Medium Intervention Subgroup	Low Intervention Subgroup	-.738	.523	.340	-1.99	.51
		High Intervention Subgroup	-.429	.476	.641	-1.57	.71
	Low Intervention Subgroup	Medium Intervention Subgroup	.738	.523	.340	-.51	1.99
		High Intervention Subgroup	.310	.591	.860	-1.10	1.72
	High Intervention Subgroup	Medium Intervention Subgroup	.429	.476	.641	-.71	1.57
		Low Intervention Subgroup	-.310	.591	.860	-1.72	1.10
LLAS	Medium Intervention Subgroup	Low Intervention Subgroup	-1.509	.909	.228	-3.68	.66
		High Intervention Subgroup	.049	.827	.998	-1.93	2.03
	Low Intervention Subgroup	Medium Intervention Subgroup	1.509	.909	.228	-.66	3.68
		High Intervention Subgroup	1.557	1.027	.289	-.90	4.01
	High Intervention Subgroup	Medium Intervention Subgroup	-.049	.827	.998	-2.03	1.93
		Low Intervention Subgroup	-1.557	1.027	.289	-4.01	.90
foot_post_index	Medium Intervention Subgroup	Low Intervention Subgroup	.2321	.7810	.953	-1.635	2.099
		High Intervention Subgroup	.5357	.7105	.732	-1.163	2.234
	Low Intervention Subgroup	Medium Intervention Subgroup	-.2321	.7810	.953	-2.099	1.635
		High Intervention Subgroup	.3036	.8822	.937	-1.805	2.412
	High Intervention Subgroup	Medium Intervention Subgroup	-.5357	.7105	.732	-2.234	1.163
		Low Intervention Subgroup	-.3036	.8822	.937	-2.412	1.805
length_popangle	Medium Intervention Subgroup	Low Intervention Subgroup	-9.554*	3.991	.050	-19.09	-.01
		High Intervention Subgroup	-2.143	3.630	.826	-10.82	6.54
	Low Intervention Subgroup	Medium Intervention Subgroup	9.554*	3.991	.050	.01	19.09
		High Intervention Subgroup	7.411	4.507	.234	-3.36	18.19
	High Intervention Subgroup	Medium Intervention Subgroup	2.143	3.630	.826	-6.54	10.82
		Low Intervention Subgroup	-7.411	4.507	.234	-18.19	3.36
motskills_centile	Medium Intervention Subgroup	Low Intervention Subgroup	-54.545*	3.777	.000	-63.57	-45.52
		High Intervention Subgroup	8.929*	3.436	.030	.72	17.14
	Low Intervention Subgroup	Medium Intervention Subgroup	54.545*	3.777	.000	45.52	63.57
		High Intervention Subgroup	63.473*	4.266	.000	53.28	73.67
	High Intervention Subgroup	Medium Intervention Subgroup	-8.929*	3.436	.030	-17.14	-.72
		Low Intervention Subgroup	-63.473*	4.266	.000	-73.67	-53.28
total_qol_child	Medium Intervention Subgroup	Low Intervention Subgroup	-1.798	4.459	.914	-12.46	8.86
		High Intervention Subgroup	22.857*	4.057	.000	13.16	32.55
	Low Intervention Subgroup	Medium Intervention Subgroup	1.798	4.459	.914	-8.86	12.46
		High Intervention Subgroup	24.655*	5.037	.000	12.61	36.70
	High Intervention Subgroup	Medium Intervention Subgroup	-22.857*	4.057	.000	-32.55	-13.16
		Low Intervention Subgroup	-24.655*	5.037	.000	-36.70	-12.61
funct_6MWD_m	Medium Intervention Subgroup	Low Intervention Subgroup	-22.738	25.981	.658	-84.84	39.37
		High Intervention Subgroup	52.952	23.635	.071	-3.55	109.45
	Low Intervention Subgroup	Medium Intervention Subgroup	22.738	25.981	.658	-39.37	84.84
		High Intervention Subgroup	75.690*	29.346	.031	5.54	145.84
	High Intervention Subgroup	Medium Intervention Subgroup	-52.952	23.635	.071	-109.45	3.55
		Low Intervention Subgroup	-75.690*	29.346	.031	-145.84	-5.54
YBT_both_cm	Medium Intervention Subgroup	Low Intervention Subgroup	-7.47498	3.75143	.122	-16.4580	1.5081
		High Intervention Subgroup	2.50675	3.60062	.766	-6.1151	11.1286
	Low Intervention Subgroup	Medium Intervention Subgroup	7.47498	3.75143	.122	-1.5081	16.4580
		High Intervention Subgroup	9.98174	4.34161	.063	-.4145	20.3780
	High Intervention Subgroup	Medium Intervention Subgroup	-2.50675	3.60062	.766	-11.1286	6.1151
		Low Intervention Subgroup	-9.98174	4.34161	.063	-20.3780	.4145
msl_endurance_s	Medium Intervention Subgroup	Low Intervention Subgroup	-19.429	11.743	.229	-47.50	8.64
		High Intervention Subgroup	11.571	10.682	.527	-13.96	37.11
	Low Intervention Subgroup	Medium Intervention Subgroup	19.429	11.743	.229	-8.64	47.50
		High Intervention Subgroup	31.000	13.264	.057	-.71	62.71
	High Intervention Subgroup	Medium Intervention Subgroup	-11.571	10.682	.527	-37.11	13.96
		Low Intervention Subgroup	-31.000	13.264	.057	-62.71	.71
PA_score	Medium Intervention Subgroup	Low Intervention Subgroup	-.27572	.20666	.381	-.7697	.2183
		High Intervention Subgroup	.22385	.18801	.462	-.2256	.6733
	Low Intervention Subgroup	Medium Intervention Subgroup	.27572	.20666	.381	-.2183	.7697
		High Intervention Subgroup	.49957	.23343	.089	-.0585	1.0576
	High Intervention Subgroup	Medium Intervention Subgroup	-.22385	.18801	.462	-.6733	.2256
		Low Intervention Subgroup	-.49957	.23343	.089	-1.0576	.0585

*. The mean difference is significant at the 0.05 level.

Figure. Post Hoc analysis Tukey HSD of subgroups

Appendix 29: COREQ Checklist

COREQ (COnsolidated criteria for REporting Qualitative research) Checklist

A checklist of items that should be included in reports of qualitative research. You must report the page number in your manuscript where you consider each of the items listed in this checklist. If you have not included this information, either revise your manuscript accordingly before submitting or note N/A.

Topic	Item No.	Guide Questions/Description	Reported on Page No.
Domain 1: Research team and reflexivity			
Personal characteristics			
Interviewer/facilitator	1	Which author/s conducted the interview or focus group?	
Credentials	2	What were the researcher's credentials? E.g. PhD, MD	
Occupation	3	What was their occupation at the time of the study?	
Gender	4	Was the researcher male or female?	
Experience and training	5	What experience or training did the researcher have?	
Relationship with participants			
Relationship established	6	Was a relationship established prior to study commencement?	
Participant knowledge of the interviewer	7	What did the participants know about the researcher? e.g. personal goals, reasons for doing the research	
Interviewer characteristics	8	What characteristics were reported about the inter viewer/facilitator? e.g. Bias, assumptions, reasons and interests in the research topic	
Domain 2: Study design			
Theoretical framework			
Methodological orientation and Theory	9	What methodological orientation was stated to underpin the study? e.g. grounded theory, discourse analysis, ethnography, phenomenology, content analysis	
Participant selection			
Sampling	10	How were participants selected? e.g. purposive, convenience, consecutive, snowball	
Method of approach	11	How were participants approached? e.g. face-to-face, telephone, mail, email	
Sample size	12	How many participants were in the study?	
Non-participation	13	How many people refused to participate or dropped out? Reasons?	
Setting			
Setting of data collection	14	Where was the data collected? e.g. home, clinic, workplace	
Presence of non-participants	15	Was anyone else present besides the participants and researchers?	
Description of sample	16	What are the important characteristics of the sample? e.g. demographic data, date	
Data collection			
Interview guide	17	Were questions, prompts, guides provided by the authors? Was it pilot tested?	
Repeat interviews	18	Were repeat inter views carried out? If yes, how many?	No
Audio/visual recording	19	Did the research use audio or visual recording to collect the data?	
Field notes	20	Were field notes made during and/or after the inter view or focus group?	
Duration	21	What was the duration of the inter views or focus group?	
Data saturation	22	Was data saturation discussed?	
Transcripts returned	23	Were transcripts returned to participants for comment and/or	

Topic	Item No.	Guide Questions/Description	Reported on Page No.
		correction?	
Domain 3: analysis and findings			
Data analysis			
Number of data coders	24	How many data coders coded the data?	
Description of the coding tree	25	Did authors provide a description of the coding tree?	
Derivation of themes	26	Were themes identified in advance or derived from the data?	
Software	27	What software, if applicable, was used to manage the data?	
Participant checking	28	Did participants provide feedback on the findings?	
Reporting			
Quotations presented	29	Were participant quotations presented to illustrate the themes/findings? Was each quotation identified? e.g. participant number	
Data and findings consistent	30	Was there consistency between the data presented and the findings?	
Clarity of major themes	31	Were major themes clearly presented in the findings?	
Clarity of minor themes	32	Is there a description of diverse cases or discussion of minor themes?	

Developed from: Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *International Journal for Quality in Health Care*. 2007. Volume 19, Number 6: pp. 349 – 357

Once you have completed this checklist, please save a copy and upload it as part of your submission. DO NOT include this checklist as part of the main manuscript document. It must be uploaded as a separate file.

Appendix 30: CSTAR Statistical Analysis

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Header: TCD145-Ward

Date: February 18, 2022

Cluster analysis is a powerful statistical method for processing data. It tries to find observations which are similar such that when they are grouped together, they should be homogenous within a group, typically an observation of one group should not be quite distinct from observations in any of the other groups. The main advantage of cluster analysis is that there are no prior assumptions made about the possible relationships in the data, making prior assumptions is the main weakness of most statistical methods.

In regard to sample size, it has been argued that there can be no universal rule of thumb concerning the minimum sample size required (Dolnicar, 2002). This is the main limitation in using the cluster analysis: the method will give an answer irrespective of how many observations used. Thus to overcome this limitation and still utilize clustering, a large sample size is needed to provide powerful and trustable results.

However, Formann (1984) suggests the minimal sample size include no less than 2^k cases (k = number of variables), and preferably 5*(2^k). The following table shows how that might look for a range of numbers of variables used in cluster analysis:

Number of variables	N (per Formann rule-of-thumb)	
	Minimum	Preferred
5	32	160
7	128	640
10	1024	5120

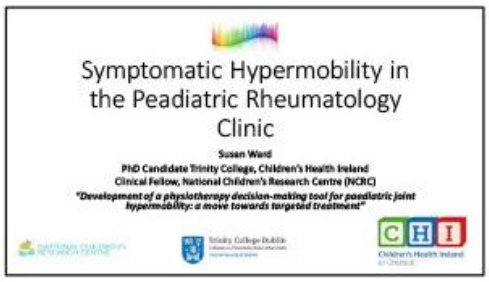
It is recommended that you reduce the number of variables in some sensible way in advance. You should ensure that the sample size is large enough to guarantee considerable clusters and generate reproducible results. It is often recommended that you split your sample, and keep a hold-out set (maybe 25% of the observations) for validation of the clusters.

References

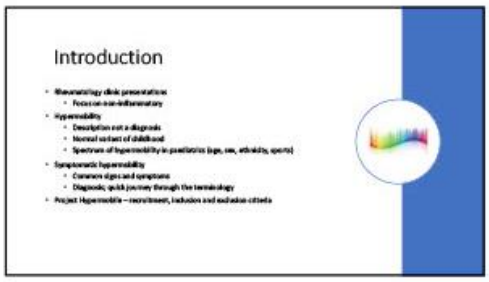
1. Formann, A. K. (1984). *Die latent-class-analyse: Einführung in Theorie und Anwendung*. Beltz.
2. Dolnicar, S. (2002). A review of unquestioned standards in using cluster analysis for data-driven market segmentation.

Appendix 31: Examples of PhD Outputs

1. Presentation Pre-Testing for CHI Rheumatology Team – 2020



1



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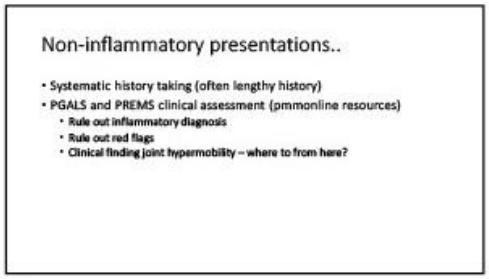
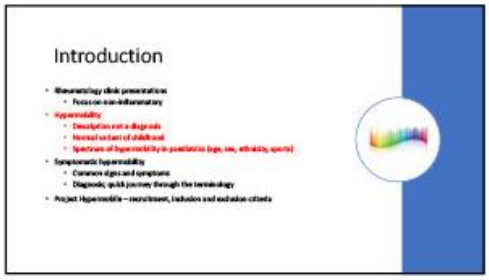
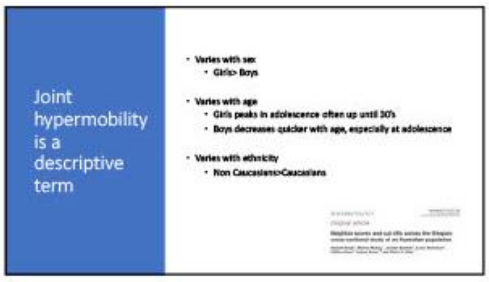


3



4



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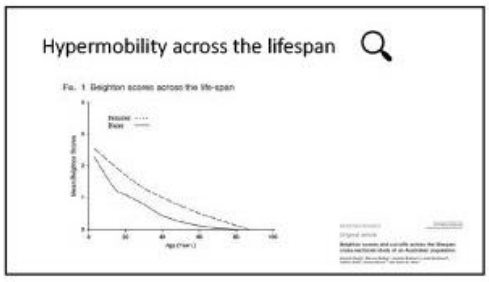

6

1

Prevalence hypermobility in children

- Irish study 20.9% prevalence
- UK 19.8% School children 13yrs
- Dutch 9.4% - 35%

- Difficult to compare studies
- Consider the assessment tool, are there standardised measures, what cut off score is appropriate for what age, is this tool valid and reliable, what joints does it cover, what does it miss?
- Identification of joint laxity, now what?

7

Introduction

- Rheumatology clinic presentations
  - Focus on non-inflammatory
- Hypermobility
  - Description not a diagnosis
  - Normal variant of childhood
  - Spectrum of hypermobility in paediatrics (age, sex, ethnicity, sports)
- Non-inflammatory symptoms with evidence of hypermobility
  - 'Symptomatic hypermobility'
  - Common signs and symptoms
  - Diagnosis; quick journey through the terminology
- Project Hypermobile – recruitment, inclusion and exclusion criteria

8

Non-inflammatory MSK symptoms in CYP (children and young people) where there is evidence of joint hypermobility

- Mechanical pain pattern (following activity, late afternoon, evening or night pain (rule out red flags))
- Stiffness, not as common, more often related to activity the day before
- Swelling – may be present but not as pronounced
- Family history – non-inflammatory (chronic pain, ?EDS, hypermobility, other diagnoses like fibromyalgia)
- Fatigue – tire more than peer group
- Poor balance or co-ordination
- 'clicking' joints
- Difficulties sitting still, poor organisation of movement
- Easy bruising
- Handwriting difficulties

9

Non-inflammatory signs and symptoms + evidence of joint hypermobility

Journey through diagnosis & terminology is confusing for medical professionals..

Imagine how it is for children and families!

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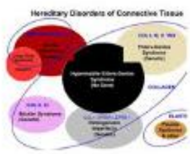

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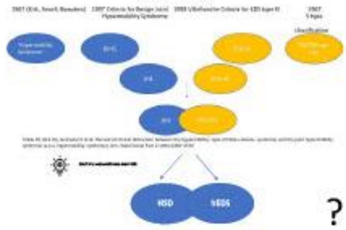
Terminology



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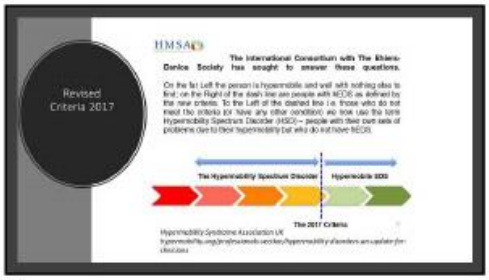

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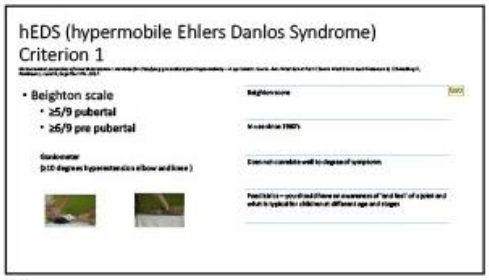

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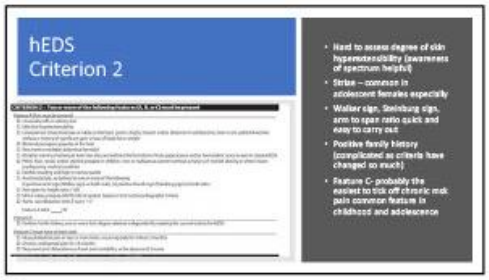

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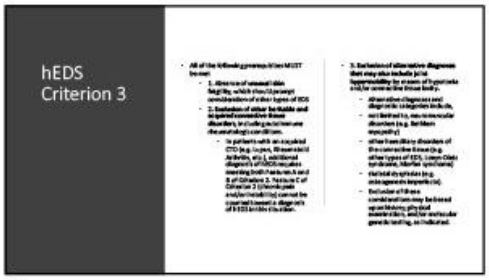

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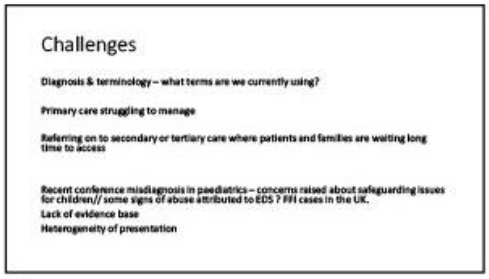

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Management CHI

- Physiotherapy led-triage clinic seeing most of the hypermobile cohort (identified by referral letter)
- Consultant clinics may see small number
- Terminology as per BSPAR recommendations 'hEDS' and 'Symptomatic Hypermobility'
- Management based on individual assessment
- Limited evidence but largely therapy led (MDT)

19

Research Hypothesis

- Children and adolescents with Symptomatic Hypermobility (Hypermobility Spectrum Disorder (HSD))or hypermobile Ehlers Danlos Syndrome (hEDS)) can be assigned to clinical subgroups using a decision-making tool based on subjective and objective clinical assessment measures.

Aim

- Develop a clinically based decision-making tool to assess children and adolescents with symptomatic hypermobility to facilitate the identification of clinical subgroups and help guide clinicians to more tailored treatment.

Objectives

- Define the clinical characteristics of new referrals (6-16 years) with a diagnosis of symptomatic hypermobility or hEDS.
- Identify subgroups of patients using cluster analysis.
- Develop a decision-making tool based on the characteristics of these subgroups.

20

Recruitment

Paediatric Rheumatology Clinic in CHI

Ethical Approval CHI and TCD

Inclusion criteria

- New referral
- Age 6 -16 years old
- Diagnosis of Symptomatic Hypermobility (features of Hypermobility Spectrum Disorder (HSD) or hypermobile Ehlers Danlos Syndrome (hEDS))

Exclusion Criteria

- Diagnosis of inflammatory disease
- Trauma or surgery that would affect functioning.
- Any other significant co-morbidities that would affect the ability to carry out the objective testing.

21

Diagnosis of Symptomatic Hypermobility

General Principles pREMS

Look

Feel

Move

Functional assessment

22

The Paediatric Regional Examination of the Musculoskeletal System (pREMS)

Assess each joint as per pGALS

pREMS details a more thorough examination in each joint

Includes assessment for hypermobility in each joint

23

Localised Hypermobility

- 4 or less joints

Peripheral Joint hypermobility

- Must have JH involving both hands and both feet
- Absence of LARGE joint or AXIAL (no arms, legs, or spine)

Generalised Joint Hypermobility

- Assessed using the Beighton Score (goniometry for elbows and knees)
- 5 or more joints (post puberty) 6 or more joints (pre puberty)
- GJH Beighton Score
- If you find generalised hypermobility you should assess for hEDS criteria

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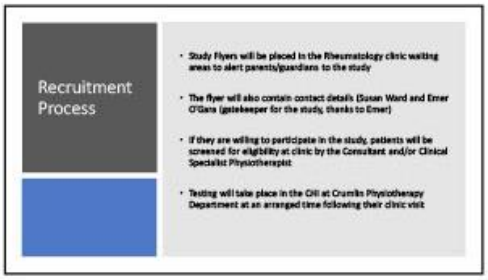
Recruitment Process
Study Flyers will be placed in the Rheumatology clinic waiting areas to alert parents/guardians to the study
The flyer will also contain contact details (Susan Ward and Emer O'Gara (gatekeeper for the study, thanks to Emer)
If they are willing to participate in the study, patients will be screened for eligibility at clinic by the Consultant and/or Clinical Specialist Physiotherapist
Testing will take place in the CHI at Crumlin Physiotherapy Department at an arranged time following their clinic visit

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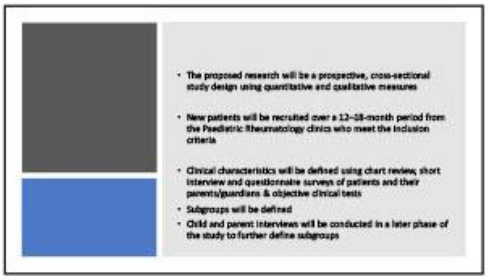
The proposed research will be a prospective, cross-sectional study design using quantitative and qualitative measures
New patients will be recruited over a 12–18-month period from the Paediatric Rheumatology clinics who meet the inclusion criteria
Clinical characteristics will be defined using chart review, short interview and questionnaire surveys of patients and their parents/guardians & objective clinical tests
Subgroups will be defined
Child and parent interviews will be conducted in a later phase of the study to further define subgroups

26

27


Medical chart review
(I will gather this information from the patient's chart, following their clinic appointment)
Diagnosis
1. hEDS use criteria
Must fit the criteria 1 (must use a goniometer (elbow and knee) when doing the Beighton) 2 and 3.
OR
2. Symptomatic hypermobile (using pGALS and pSIMS Typical rheumatology assessment)
List symptoms
List parent or child report of joints experiencing pain for 3 months or greater
Medications
List number of days missed 'schooling' (attendance at school or engagement with schooling at home) related to symptoms

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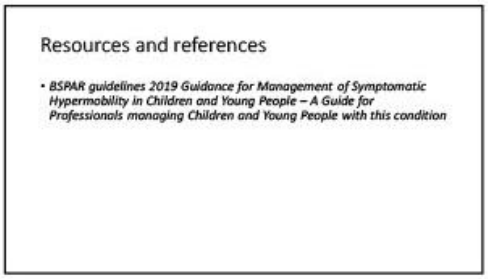
Resources and references
BSPAR guidelines 2019 Guidance for Management of Symptomatic Hypermobility in Children and Young People – A Guide for Professionals managing Children and Young People with this condition

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2. Presentation for CHI Rheumatology Team – 2021

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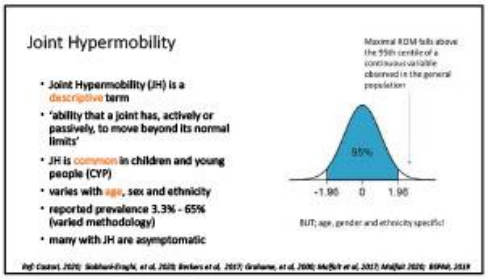

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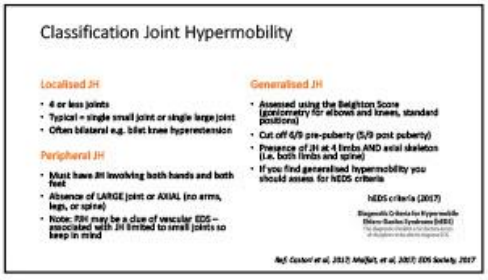

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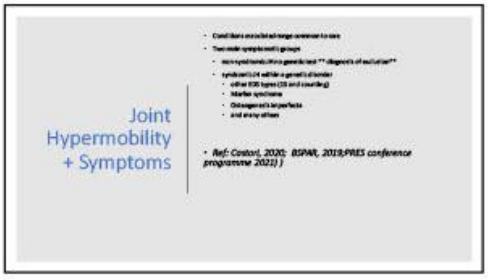

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Journey through the acronyms…

1967 1988 2000's 2017 2021?

HMS BJHS EDS III JHS EDS-HT HS hEDS HSD symptomatic hypermobility

Still used (FREQ)

Terms in blue = recognised by BSPAR, position statement July 2019

Ref: Castori, 2010; BSPAR, 2019;PAES conference programme 2021

7

Symptoms

- Marked heterogeneity
- *"overlapping pool of vague subjective symptoms"*
- Difficult to interpret
- Variation
  - Age
  - Gender
  - Diagnostic criteria
  - Outcome measure
  - Clinical setting
  - Study design

Some of the (many) clinical characteristics associated with JH in the literature

Ref: Kala, 2010; Simanek, et al. 2010

8

Scoping review objectives

- Map the literature
- Across all clinical domains
- Children and young people 0-24 years
- Use internationally recognised diagnostic terminology
- Identify clinical characteristics of this heterogenic cohort

9

Research Question

"What are the presenting clinical characteristics of children and young people diagnosed with symptomatic hypermobility (including HSD and hEDS and other equivalent diagnoses)?".

10

Study Design

- Scoping review (ScR) methodology
- Systematic approach to explore literature, mapping and summarising the evidence available
- Provides an overview of the evidence
- ScR allows for a broad inclusion criteria to answer the research question in a comprehensive manner
- Reported in line with Preferred Reporting Items for Systematic Reviews and Meta-Analyses extension for scoping reviews (PRISMA-ScR)
- ScR is an iterative process – allowing for clarification, discussion and amendment of process as reviewers progress through the ScR

Ref: Arksey and O'Malley, 2005

11

Criteria for inclusion

Population

- Children and young people age 0-24 years (overlaps WHO definition of adolescence 10-19 and youth 15-24)
- Confirmed clinical diagnosis of symptomatic hypermobility (2017 hEDS and HSD)
- Including previous terminology (JHS/BJHS, EDS-HT/EDS III)
- Studies including asymptomatic JH (the trait) will be omitted

Concept

- Map clinical characteristics across all medical domains
- Clinical assessment must be conducted using documented outcome measures

Context

- Including RCT, observational and interventional studies, reviews, case reports, book chapters, grey literature searches, consulting with experts, websites, handsearching of key articles and reviews

Ref: Peters, et al, 2020.

12

Identifying relevant studies

- EMBASE, Medline, CINAHL, and Web of Science
- Search extended to grey literature, conferences, websites, consultation with experts, manual searching of targeted websites
- Review articles hand searched to identify articles not yet captured

13

Study selection

- Identified studies exported to Endnote software
- Duplicates removed and exported to Covidence software
- Two stages
  - Title and abstract screening
  - Full text review
- Two independent reviewers apply criteria to all citations and subsequently to full text
- Conflicts are resolved by a third reviewer

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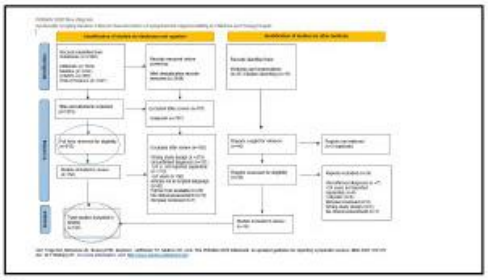

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Preliminary Results

- The search identified 1619 studies published from 1967 to 2021
- 164 articles were included (n=4554).
- Diagnostic criteria, outcome measures, and study settings were highly heterogenous.
- The main study designs were reviews (32%), case reports (24%) and cross sectional (21%) and 1.2% were longitudinal.
- The Beighton Score was frequently used (49%), but rarely standardised (7%).
- Outcome measures varied with clinical setting.
- There was a wide variation in clinical characteristics reflecting 17 different clinical domains.
- Musculoskeletal symptoms were the most prevalent in all age groups
- Domains such as cardiovascular were associated with specific age groups in particular early adolescence to young adulthood.

16

17

Irish Childhood Hypermobility Project

Inclusion Criteria

Inclusion criteria

- New referral
- Age 6 -16 years old
- Diagnosis of Symptomatic hypermobility or hypermobile Ehlers Danlos Syndrome (hEDS)

Exclusion Criteria

- Diagnosis of inflammatory disease
- Trauma or surgery that would affect functioning.
- Any other significant co-morbidities that would affect the ability to carry out the objective testing.

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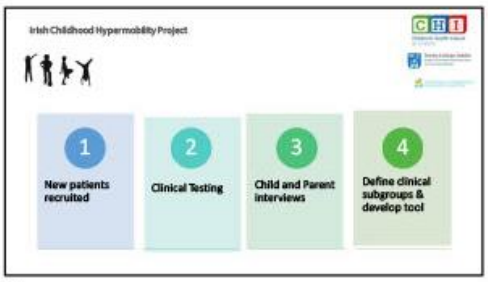
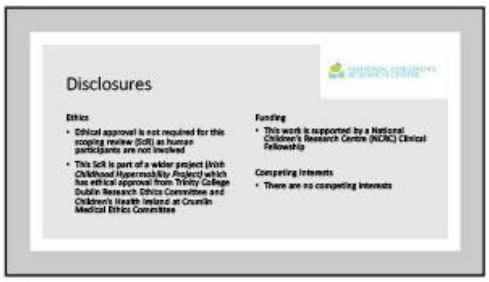

Irish Childhood Hypermobility Project
1
New patients recruited
2
Clinical Testing
3
Child and Parent Interviews
4
Define clinical subgroups & develop tool

19


Disclosures
Ethics
Ethical approval is not required for this scoping review (ScR) as human participants are not involved
This ScR is part of a wider project (Irish Childhood Hypermobility Project) which has ethical approval from Trinity College Dublin Research Ethics Committee and Children's Health Ireland at Crumlin Medical Ethics Committee
Funding
This work is supported by a National Children's Research Centre (NCRC) Clinical Fellowship
Competing Interests
There are no competing interests

20


With thanks
Research Team
Dr Emma MacDermott, Consultant Paediatric Rheumatologist, Children's Health Ireland at Crumlin
Dr Sara Dockrell, Assistant Professor, Discipline of Physiotherapy, Trinity College Dublin
Dr Jane Simmonds, Associate Professor from Great Ormond Street Institute of Child Health, University College London
Dr Janet Deane, NIHR Clinical Lecturer and AHP Research and Innovation Lead, University of Manchester and Manchester University NHS Foundation Trust
David Mockler, Assistant Librarian Reader Services, Library Trinity College Dublin
Special thanks
Rheumatology Team OH at Crumlin/at Blanch
Dr Orla Killeen
Edel Carberry
Wider team for support in recruitment for Irish Childhood Hypermobility Project

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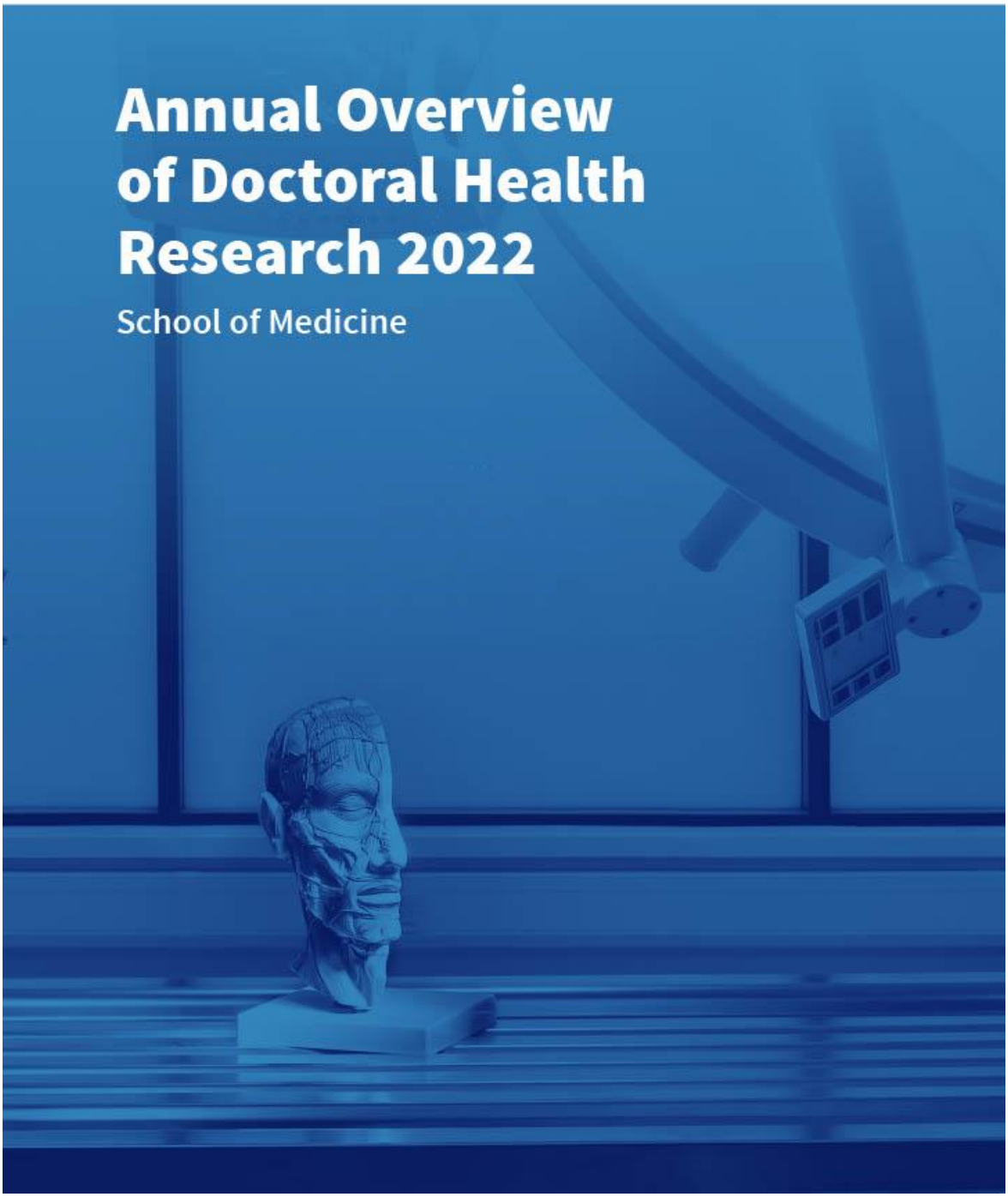
3. Annual Overview of Doctoral Health Research 2022 – School of Medicine, TCD

Trinity College Dublin
Coláiste na Tríonóide, Baile Átha Cliath
The University of Dublin



DISCIPLINE: PHYSIOTHERAPY

Symptomatic hypermobility in children: a move towards improved care pathways and targeted treatment

PhD Student: Susan Ward
Supervisor: Dr Sara Dockrell and Dr Emma McDermott
Funder: National Children's Research Centre

Overview of Project:
Susan is researching children with joint hypermobility-related disorders. Joint hypermobility is the term used to describe the capability that a joint has to move beyond normal limits. For most children, hypermobility causes no problem. However, some children complain of pain in multiple joints and muscles, frequently worse towards the end of the day following activity. They may complain of fatigue or difficulties with functional tasks which can limit their ability to participate in daily activities (such as attending school or doing sport and physical activity). Symptoms often extend beyond the musculoskeletal system into the gastrointestinal, cardiovascular, urogenital, and psychological domains. This varied pattern of symptoms can present a challenge and delays in terms of diagnosis and treatment. Exploring this range of symptoms in children is an important part of Susan's mixed methods research to help us understand the condition and the full impact of it on the children's quality of life. Susan intends to assess the clinical characteristics in participants diagnosed with symptomatic hypermobility using a cross-sectional quantitative study. Additionally, semi-structured interviews will be used to explore the impact of symptomatic hypermobility on children and their families. She hopes to develop a decision-making tool for use in the clinical setting.

Long-term impact of project:
The team hope to inform and influence future care pathways for children with symptomatic hypermobility who currently experience long delays in diagnosis and management.

Quote:
"Following my PhD, as a research active health professional, I hope to encourage others to engage in meaningful research in their own clinical areas of expertise."

4. Poster for launch of Annual Overview of Doctoral Health Research 2022 – School of Medicine, TCD

Paediatric Symptomatic Hypermobility: A Move Towards Targeted Treatment

Susan Ward[1]; Sara Dockrell[1]; Emma MacDermott[2]; Janet Deane[3]; Jane Simmonds[4]
[1]Discipline of Physiotherapy TCD, [2] Children's Health Ireland, [3] University of Birmingham, [4] University College London

Background

Joint Hypermobility (JH) is present when a joint actively or passively moves beyond its normal range of movement[1]. JH is represented by a continuum and varies with age. It also varies with sex, ethnicity, strength training or previous injuries, which is challenging for the clinician[2]. Symptomatic JH is a term used to define symptoms thought to be related to JH. Children and young people typically present with joint pain but can also present with symptoms across many other clinical domains, often mimicking other conditions which may be common in the general paediatric population[3]. This varied pattern of symptoms means that delays to treatment are common. Defining more homogenous subgroups could help to improve and target treatment[4].

Methods

A scoping review using PRISMA methodology was conducted. Full text articles reporting on children and young people from birth to 24 years with a confirmed diagnosis of symptomatic hypermobility were included.
A prospective cross-sectional study was carried out on children and young people diagnosed with symptomatic hypermobility, age 6-16 years attending the rheumatology service in a tertiary children's hospital. Medical charts were reviewed, and a structured interview and clinical measurements were conducted including assessments of joint hypermobility, function, balance, strength and endurance, posture and gross and fine motor skills (Figure 1). Assessments of pain, quality of life, multidimensional fatigue and physical activity were also carried out using standardised tools.

Figure 1. Clinical Measurements Y-Balance Test and Fine Motor Skills

Results

The scoping review including 163 studies. Clinical characteristics varied with developmental stage (Figure 2). Inconsistent diagnostic criteria and assessment methods were identified (Table 1).

Eighty children (26 males, 54 females) were included in the cross-sectional study. Mean age was 11.57(+/-3.0) years (range 6.0-16.92). All participants reported pain in more than 1 joint for greater than 3 months, most affected were the knee (72.5%) and ankle (55%) joints. The majority (87.5%) reported more than one symptom. The most common clinical presentation at onset was joint pain or swelling (n=55, 68.%). Other musculoskeletal presentations included flat feet, the presence of hypermobile joints and the occurrence of frequent falls or repeated injuries. Non-musculoskeletal issues included motor delay or difficulties, persistent fatigue and gastrointestinal issues (Chart 1). Below average motor skills were identified in 44% of participants.

Figure 2. Percentage of studies for each developmental stage reporting clinical characteristics grouped in clinical domains

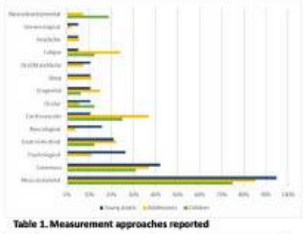


Table 1. Measurement approaches reported

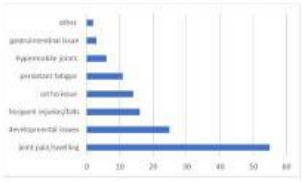


Chart 1.Clinical Presentation at onset

Discussion

This is the first scoping review to explore literature on children and young people with symptomatic hypermobility with a unique focus on development. Whilst musculoskeletal symptoms are the most prevalent, clinical characteristics were reported across 14 different clinical domains. In adolescence, dysautonomia, fatigue and sleep issues were more common, whilst psychological characteristics were reported in over a quarter of all young adult studies. The variety of measurement approaches used may represent a growing awareness of the spectrum of presentations but could also indicate the lack of guidance and use of specific standardised measurements.
Children and young people presenting to a tertiary paediatric rheumatology service predominantly present with chronic joint pain but also present with a spectrum of other musculoskeletal and non-musculoskeletal complaints. Motor skills were poor and may not be suitably targeted by current treatment approaches.

Conclusions

Scoping Review. There is a changing phenotype of symptomatic hypermobility from childhood to young adulthood. There is a lack of guidance regarding the use of standardised tools. There is a need for defined, bespoke criteria and assessments.

Cross-sectional Study. This study identified the high frequency of multisystem involvement. Motor skill proficiency is poor which will impact on participation in sports and activities. A holistic approach to assessment is required to recognise the clinical significance of JH including assessment of quality of life, fatigue and motor skill acquisition.

Gaining clarity to this heterogenous condition, may help management of patients with symptomatic hypermobility which is currently based on a limited evidence base[5]. Earlier recognition, followed by a targeted care pathway, may improve management for children and young people with symptomatic hypermobility.

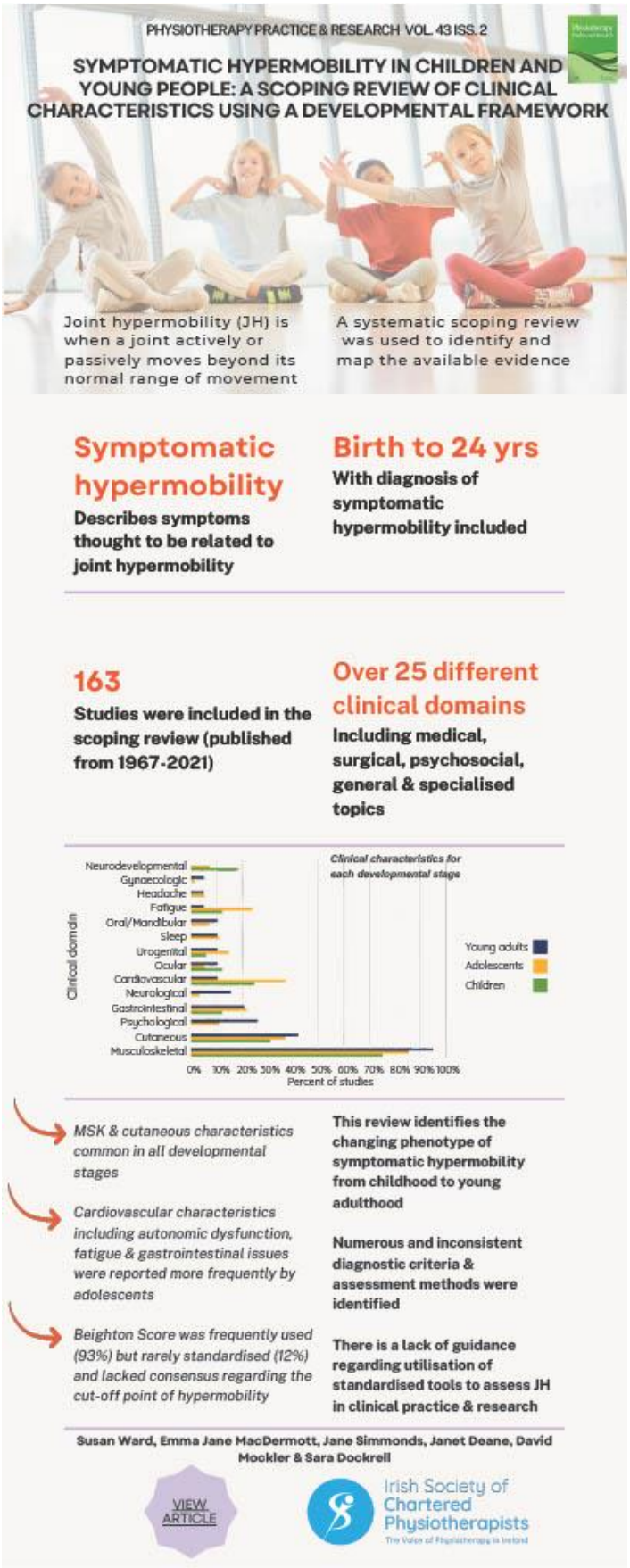
Contact

Susan Ward
Discipline of Physiotherapy
Email: wardsu@tcd.ie

References

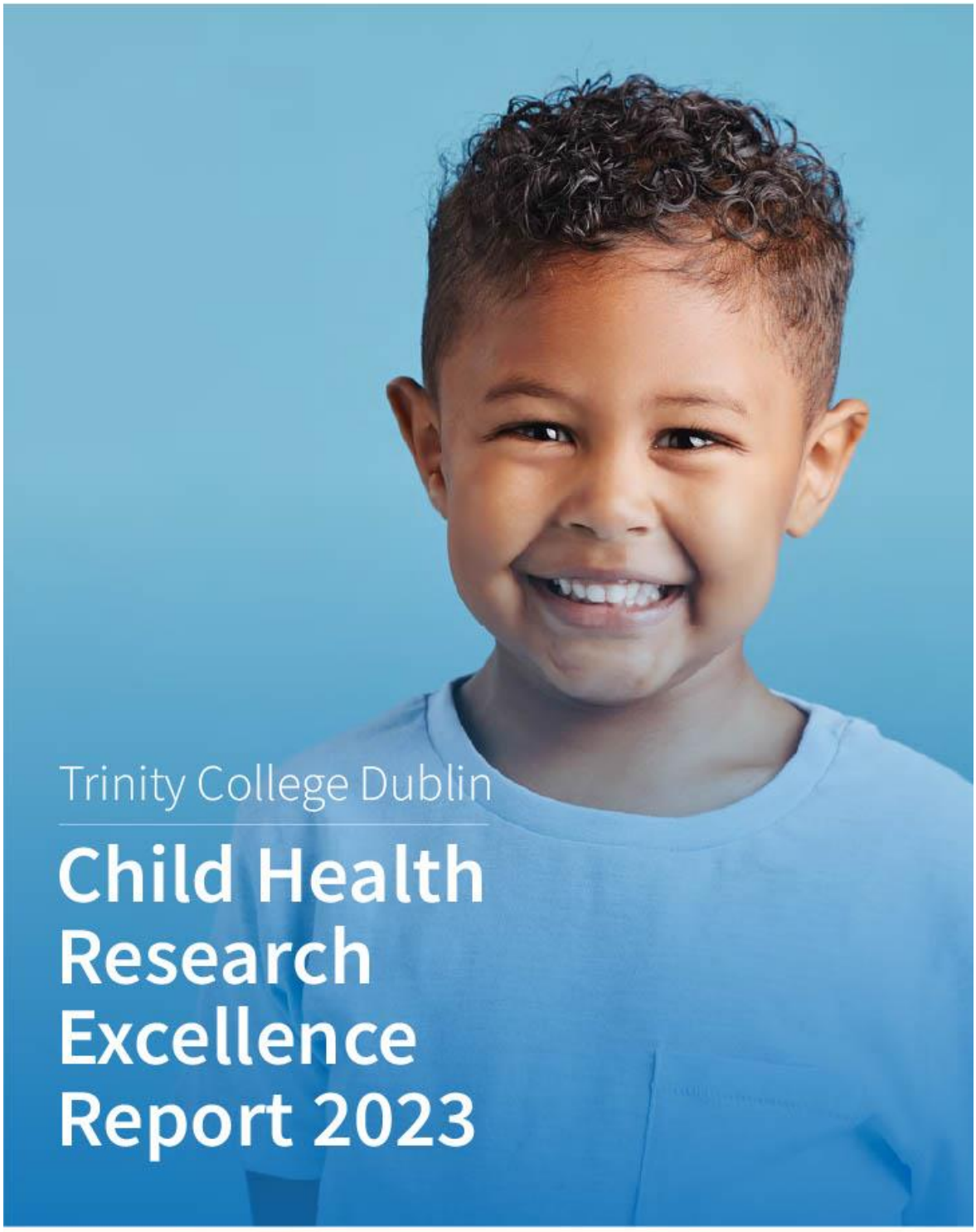
1. Tinkle BT. Symptomatic joint hypermobility. Best Practice & Research in Clinical Rheumatology. 2020;34(3):101584.
2. Juul-Kristensen B, Schmedling K, Rombaut L, Lund H, Engelbert RH. Measurement properties of clinical assessment methods for classifying generalized joint hypermobility-A systematic review. Am J Med Genet C Semin Med Genet. 2017;175(1):116-47.
3. Arnon E. Musculoskeletal pain and hypermobility in children and young people: is it benign joint hypermobility syndrome? Archives of Disease in Childhood. 2015;100(1):2-3.
4. Di Mattia K, Fary R, Murray KJ, Howie E, Smith A, Morris S. Two subtypes of symptomatic joint hypermobility: a descriptive study using latent class analysis. Archives of Disease in Childhood. 2019;104(11):1049-101.
5. Engelbert RH, Juul-Kristensen B, Pacey V, de Wandele I, Simeoni S, Wolvarosky N, et al. The evidence-based rationale for physical therapy treatment of children, adolescents, and adults diagnosed with joint hypermobility syndrome/hypermobile Ehlers Danlos syndrome. American Journal of Medical Genetics Part C, Seminars in Medical Genetics. 2017;175(1):158-67.

5. Infographic for the Irish Society of Chartered Physiotherapists (online magazine -2022)



6. Paediatric Rheumatology European Association Certificate of Membership

7. TCD - Child Health Research Excellence Report 2023



Trinity College Dublin
Coláiste na Tríonóide, Baile Átha Cliath
The University of Dublin

Childhood symptomatic hypermobility: a move towards improved care pathways and targeted treatment

Dr Sara Dockrell, Assistant Professor (Physiotherapy), School of Medicine

The key direction of Sara Dockrell's research is the investigation and promotion of musculoskeletal health and well-being in both children and student populations in education, and adults in the workplace, with an emphasis on ergonomics.

"The long term expected impact is to improve the symptomatic hypermobility patient journey for children and young people from diagnosis to treatment."

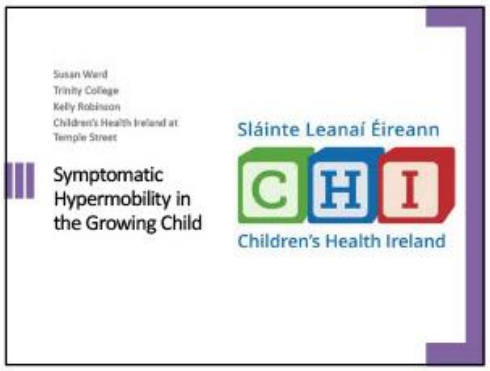
Childhood symptomatic hypermobility: a move towards improved care pathways and targeted treatment.

Joint hypermobility is the term used to describe the capability that a joint has to move beyond normal limits. Other terms used to describe this are joint laxity or double-jointedness. Children are more hypermobile than adults. For most children, their hypermobility causes no problems, and can be an advantage in some sports like gymnastics, or swimming. Symptomatic Hypermobility is a term used to describe the symptoms associated with joint hypermobility. Synonymous terms include hypermobile Ehlers Danlos Syndrome and Hypermobility Spectrum Disorders. Children and young people with symptomatic hypermobility frequently complain of pain in multiple joints and muscles which is often worse towards the end of the day or following activity. They may complain of fatigue or difficulties with functional tasks, which can limit their ability to participate in daily activities like attending school or doing sport and physical activity.

A range of gastrointestinal, cardiovascular, psychological, and urogenital characteristics are also reported to be associated with Symptomatic Hypermobility. However, varied terminology across different clinical settings makes clinical interpretation difficult in children and young people.

The diagnosis and management of children and young people with symptomatic hypermobility can be challenging. Currently, there is no consensus about how to effectively manage children and young people with joint hypermobility-related conditions. Chartered Physiotherapist Susan Ward is currently undertaking a PhD to explore: (i) the symptomatology of SH using a developmental framework; and (ii) the impact of symptomatic hypermobility on quality of life for the patients and their families. She also aims to develop a decision-making tool that could be used as part of an improved pathway for the management of patients with hypermobility-related conditions.

8. Presentation – CHI Conference Temple St Hospital, May 2023



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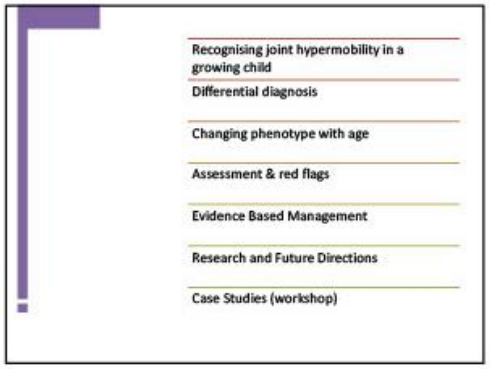

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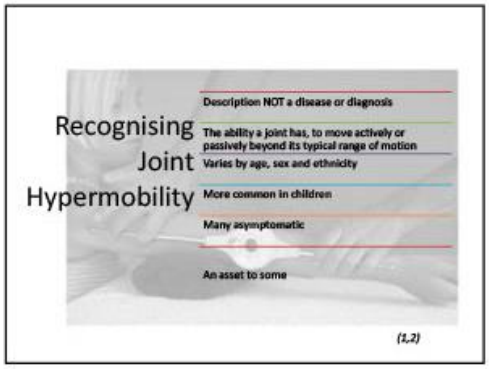

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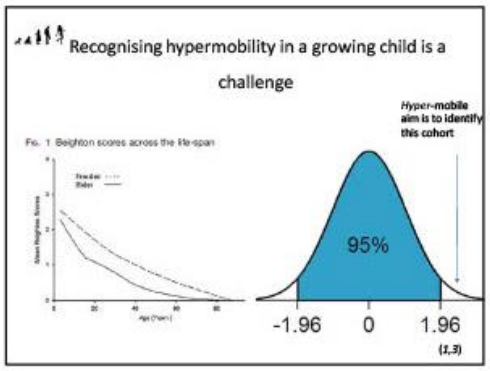

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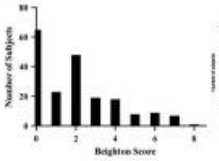
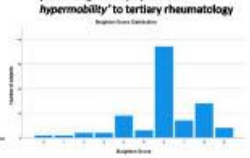
Beighton score distribution among a North American paediatric orthopaedic population n=202, 54% F, age 2-18yrs



Overall prevalence generalised JH (≥5/9)13% *(4)*

Beighton score distribution among Irish children, n=80, 65% F, 6-16years presenting with 'symptomatic hypermobility' to tertiary rheumatology



Overall prevalence generalised JH (≥ 5/9) 78.3%
≥ 6/9 - 74.7%
(unpublished data)*(5)*

Prevalence generalised JH
Primary Care Physiotherapy 21.9%
Primary school group 17.1% *(6)*

7

Key Points

Hypermobility **changes** with age

Ask what is the **clinical significance** of the hypermobility?

Avoid under or over recognition of *hypermobility*

8

1. Recognising hypermobility in a growing child
2. Differential diagnosis
3. Changing phenotype with age
4. Assessment/Red Flags
5. Evidence Based Management
6. Research and Future Directions
7. Case Study

9

Differential diagnosis..... Child presenting with joint pain

Inflammatory disease

Presenting Joint pain
- Inflammatory Pattern
- Can be unilateral or bilateral
- Often improves with activity
- Worse with rest (car journeys, sitting in class)

- Morning stiffness > a few minutes
- Persistent swelling
  - lasting more than 6 weeks
- Loss of joint range
  - can be subtle*(may also be hypermobile)
- Bony overgrowth
  - sign of chronic inflammation

Non-inflammatory

- Presenting Joint pain
  - Mechanical Pattern
  - Often bilateral
  - Pain often presents later afternoon/evenings
  - Episodic, activity related pain
  - Improves with rest
- No significant morning stiffness
- Intermittent swelling
- No loss of joint range
- Examine for joint *hypermobility*
- Rule out/exclude other causes of joint pain...

See 'Is musculoskeletal history and examination so different in paediatrics?' (7)
www.pmmonline.org *(8)*

10

Differential Diagnosis..... Paediatric Joint hypermobility

- Heritable Connective Tissue Disorders
- Rare Ehlers-Danlos syndrome (EDS) *(9)*
  - Vascular EDS 1:50,000
  - Family history
  - Congenital features
  - Skin usually a give-away at early age (significant bruising)
  - Not the most hypermobile (peripheral small joints only often)
  - Later in childhood early varicose veins, characteristic facial features
  - Challenge to diagnosis in early childhood
- Global developmental delay
- Significant Hypotonia or muscle weakness
  - joint hyperlaxity +contractures
- Non-familial features
- 'other' features
  - Facial (specific facial features)
  - Stature (short stature, other skeletal features)
  - Developmental (learning difficulties)

11

Differential Diagnosis

JH within a genetic disorder *(10)*

Some examples
- Marfan Syndrome (1:5,000-1:10,000)
  - Know the Signs - Marfan Foundation *(9)*
  - Not infrequently muscle weakness/muscle hypoplasia, typical features
- Osteogenesis Imperfecta (1:15,000-20,000)
- Classical EDS (cEDS 1:10,000)

Classical EDS vs hypermobile EDS *(11,12)*

12

The terminology of '*Symptomatic* Hypermobility'

Rheumatology

Hypermobility Syndrome (HMS)

↓

Benign Hypermobility Syndrome (BJHS)

↓

Joint Hypermobility Syndrome (JHS)

↓

BSPAR Symptomatic Hypermobility

↓

Hypermobility Spectrum Disorder (HSD)

Dermatology/Genetics

EDS-Type 3 or III

↓

Ehlers-Danlos Syndrome Hypermobility Type (EDS-HT)

↓

Hypermobile-EDS (hEDS)

↘

New! Peadiatric Diagnostic Framework; May 2023 (13)

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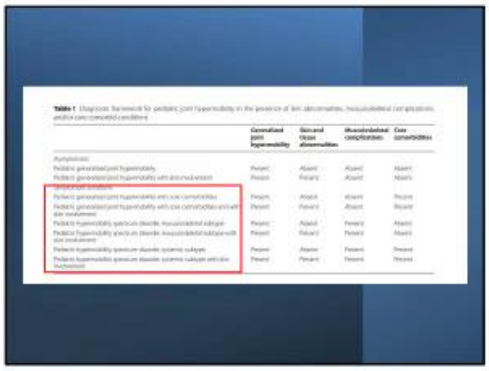

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16

Key Points

- Red Flags
- Differential diagnosis appropriate onward referral
- Some conditions don't develop their features until later in childhood

17

1. Recognising hypermobility in a growing child is a challenge!
2. Differential diagnosis – conditions with JH as a feature
3. Changing phenotype with age
4. Assessment
5. Evidence Based Management
6. Research and Future Directions
7. Case Study

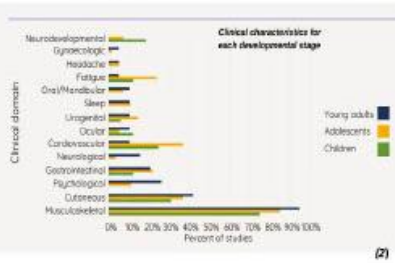
18

Changing phenotype with age

(2)

19

Changing phenotype with age



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A changing phenotype

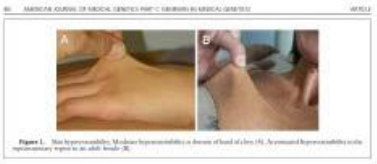
Cutaneous Features (*11,12*)

- Classical EDS can present similarly to hEDS especially in earlier childhood

Look out for

- Soft/doughy
- Extensible/stretchy
  - Assess for extensibility at neck and inside forearm
- Undress your patient to shorts/tee-shirt
- Observe shins
- Check for shin discoloration
- Scars – keloid/atrophic
- Unexplained stretch marks

21



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A changing phenotype

Musculoskeletal Features

- Joint instability is more common in early puberty onwards
- If present in childhood, consider differential diagnosis
  - Bone dysplasia,
  - Other form of EDS

23

A changing phenotype

Childhood

- Activity related joint pain
- Frequent trips and falls
- Gross motor and fine motor difficulties
- Late childhood – may have onset cutaneous features

Adolescence

- Activity related pain – more persistent in some
- Fatigue
- Changing activity levels/type
- Onset other multisystem symptoms
- Worsening or improvement of symptoms – complex interaction of events (puberty/environment/biological sex)
- Increased number of co-morbidities

(5)

24

1. Recognising hypermobility in a growing child is a challenge!
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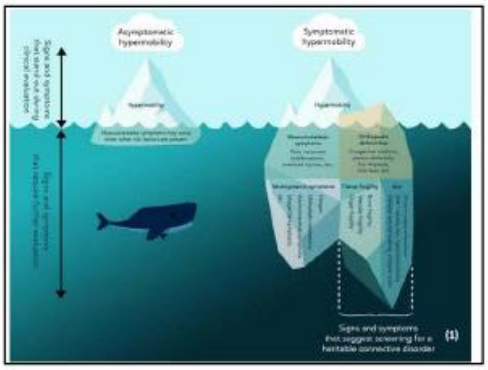

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'Clinically Relevant'

(2)

27

Proposed Assessment

Subjective & Objective – consider the following;

- Differential diagnosis & Red Flags
- Screen hypermobility and skin
- MSK laxity/strength/endurance/length
- Balance & proprioception
- Pain/Fatigue/QOL
- Functional capacity
- Motor skill
- Physical Activity
- Screen co-morbidities

28

Hypermobility screening

Beighton Score *(14,15)*

- Valid and Reliable
- Care with positioning
- Goniometry at elbows and knees
- Proposed 5-18-years-old*
- New cut off ≥ 6/9

Lower Limb Assessment Scale *(16)*

- 12 items
- Right and left lower limb
- Lower Limb hypermobility & laxity
- Multidirectional
- Cut off ≥ 7/12

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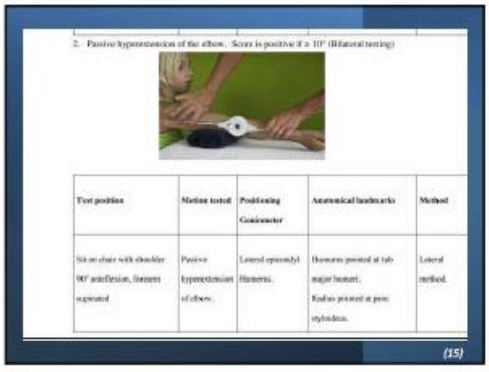

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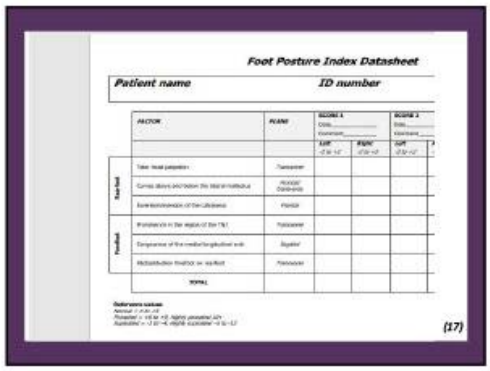

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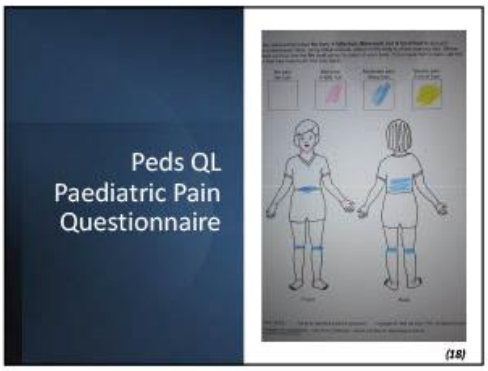

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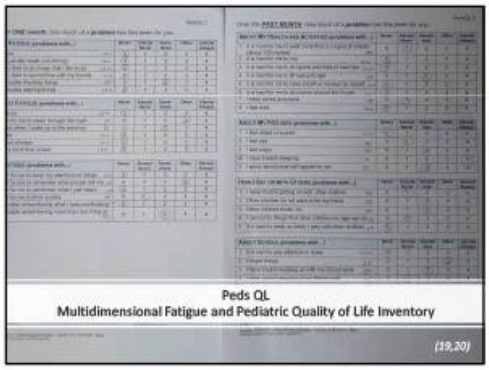

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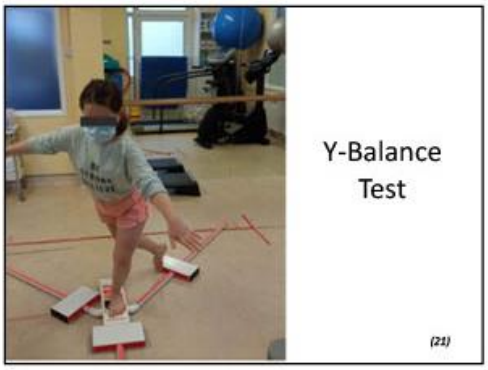

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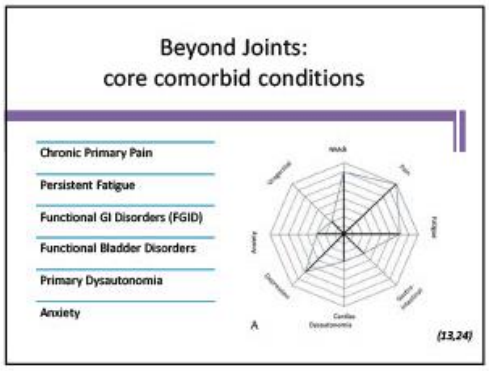

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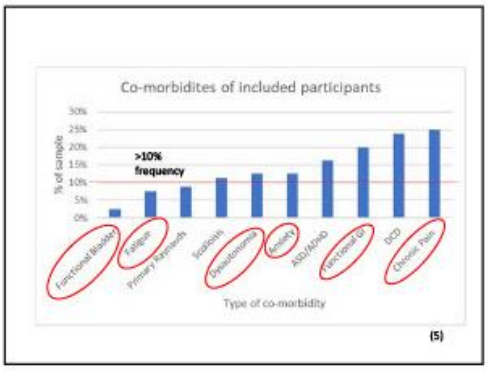

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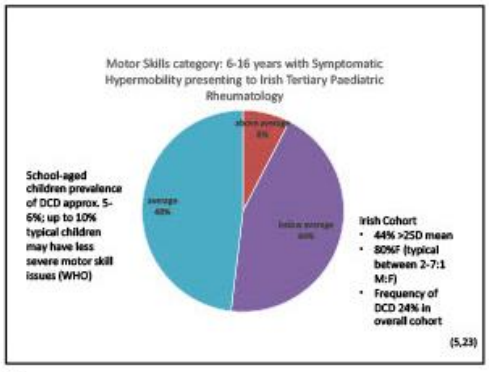

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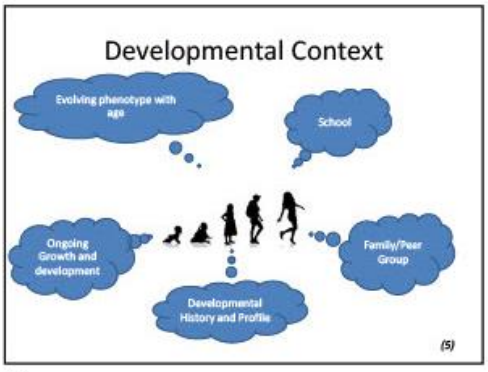

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Assessment Tips

- First contact time can be lengthy
- Assessment; rule out red flags; consider JH-related disorders (diagnosis of exclusion); beyond joints approach; developmental context
- Hypermobility screening; use a standardised protocol for Beighton; consider LLAS
- High risk? Poor function and high report of multisystem complaints (25)

43

Key Points

Beighton + goniometry + standardised positioning proposed ≥6/9 cut-off

Developmental context

Beyond joints

44

1. Recognising hypermobility in a growing child is a challenge!
2. Differential diagnosis – conditions with JH as a feature
3. Changing phenotype with age
4. Assessment
5. Evidence Based Management
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45

Evidence Based Management

ARTICLE

The Evidence-Based Rationale for Physical Therapy Treatment of Children, Adolescents, and Adults Diagnosed With Joint Hypermobility Syndrome/Hypermobile Ehlers Danlos Syndrome

RAOUL H.H. ENGELBERT, BIRGIT JUUL-KRISTENSEN, VERITY PACEY, INGE DE WANDELE, SANDY SMEENK, NICOLETA WOINAROSKY, STEPHANIE SABO, MARK C. SCHIEPER, LESLIE RUSSEK, AND JANE V. SIMMONDS

(25)

46

Evidence Based Management

British Society for Rheumatology

Guidance for Management of Symptomatic Hypermobility in Children and Young People – A Guide for Professionals managing Children and Young People with this condition

This guidance document has been compiled by the Allied Health Professionals Group working within BSPAR Section Council and has been designed to help and support therapists working with children and young people (CYP) presenting with symptomatic hypermobility and musculoskeletal pain.

The WHO definitions: Childhood 0-10 years, Adolescence 10-19 years and Young Adulthood 20-25 years.

This guidance replaces the former guidance from Jan 2013, ratified by the BSPAR Executive Committee in June 2013. In order to cover the complexities of this condition in some CYP, some aspects of management have been divided into different professions; however, there will be significant overlap as to who provides the intervention depending upon local teams.

(27)

47

Group-Based Management

CHI at Temple Street

- 6/52 x 1 hour group
- Goal focused
- Physio-led
- Graded and paced strengthening
- Cardiovascular; graded approach in a supported and fun environment
- Reassessment and support to DC locally
- Access to a virtual group for older children and teens
- Evidence of improved strength following 6/52 intervention

CHI at Crumlin

- 6/52 x 1.5 hour group
- Goal focused
- MDT-led (PT,OT, Psych and Social Work)
- Strength and fitness
- Fine motor skills programme
- Activity timetable and pacing education
- Pain Education provided to parents
- Reassessment and support to DC to local service
- Previously mixed inflammatory/hypermobility heterogenous cohort

48

Key points

EARLY RECOGNITION
KNOWLEDGEABLE CLINICIANS
EFFECTIVE COMMUNICATION
PARTNERSHIP APPROACH

- Patient centered goals
- Focus improving
  - Strength
  - Proprioception/body awareness
  - Breath control
  - Motor Skills & Physical Activity Levels
  - Knowledge of Pain Science
  - Pacing
  - Sleep hygiene
- Appropriate onward referral
- MDT where appropriate

(28)

49

Research and Future Directions

- New paediatric criteria
- Develop local expertise
- Clearly defined care pathways
- Ongoing research
  - Population studies
  - Longitudinal studies
  - Paediatric specific intervention studies
  - Standardized measurements/outcome/terminology

50

Susan Ward*
PhD candidate
Discipline of Physiotherapy Trinity College wardsu@tcd.ie
*Anyone interested in collaborating in future research with this population please reach out!

Kelly Robinson
Senior Physiotherapist
CHI at Temple Street
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51

References

[illegible]

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References

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53

9. European Paediatric Rheumatology Conference 2023 Abstract

Susan Ward European Paediatric Rheumatology Conference 2023 Abstract

Word character count 2976 (<3000 including title)

Title: Mapping clinical characteristics in children and adolescents with symptomatic hypermobility in tertiary paediatric rheumatology settings. An Irish perspective.

Introduction. Determining clinically significant joint hypermobility in childhood presents a challenge to clinicians. Previous research has demonstrated an evolving phenotype whereby hypermobility and symptoms may further develop or resolve over time. This is the first study to map clinical characteristics of children and adolescents presenting with symptomatic hypermobility in tertiary rheumatology settings from an Irish perspective.

Objectives. Define the clinical characteristics of children and adolescents presenting with symptomatic hypermobility to tertiary paediatric rheumatology services in Ireland.

Methods. A prospective cross-sectional study was carried out on children and adolescents aged 6-16 years recognised as having symptomatic hypermobility determined by a paediatric rheumatologist. A structured interview and clinical measurements were conducted. Standardised assessments included screening for generalised hypermobility (Beighton score and Lower Limb Assessment Score (LLAS)), Foot Posture Index, the 6-minute walk test, Y-balance test, measurement of strength and endurance and motor skills screening (BOT-2 Brief). Parent and child questionnaires assessing pain, multidimensional fatigue and quality of life were also completed following strict protocols.

Results. Eighty participants (26 males, 54 females) were included. Mean age was 11.6±3 years (range 6.0-16.92). Generalised joint hypermobility (Beighton score of ≥6/9) was identified in 79% of participants, and a further 9% classified using the LLAS. The mean time from onset of symptoms to recognition of symptomatic hypermobility was 32±23 months (range 3-108 months). All participants reported pain in more than one joint for >3 months; most affected were the knee (73%) and ankle (55%). Comorbidities were common with 49% reporting three or more. The most common were neurodevelopmental (48.8%) and chronic pain (25%). Below average motor skills were identified in 44% of participants. Muscle strength and endurance were poor with only 24% and 32% achieving within the normative range. Feeling tired 'often' or 'almost always' was reported by 46% of participants and finding it hard to keep attention 'often' or 'almost always' was reported by 30%. PedsQL generic core scores (60 ±18.22) were reduced compared to normative data (83±14.79).

Conclusions This study identified long delays to recognition of symptomatic hypermobility and the significant burden of symptoms and comorbidities in children and adolescents. The high levels of neurodevelopmental diagnoses, poor motor skills and multidimensional fatigue are of particular concern in this population. Earlier recognition and standardised assessments within a biopsychosocial framework are needed to develop better care pathways to improve outcomes.

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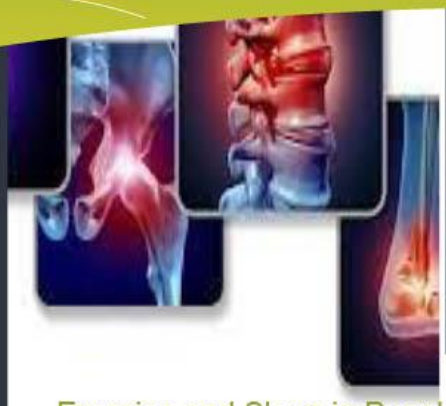
10. IRHPS Summer Newsletter 2023 Pages1&2

June 2023

IRHPS Summer Newsletter



Highlights in this issue

Autumn Annual Conference

The annual IRHPS face to face conference will be held alongside the Irish Society of Rheumatology (ISR) Autumn Meeting at Fitzpatrick's Castle Hotel, Killiney in Dublin on 21st September 2023. ISR will be a two day event 21st & 22nd September.

Registration is not yet open

The Theme of the 2023 annual IRHPS conference is "Working Together"

We Would like to invite those who are paid-up members for the last two years to submit abstracts for the IRHPS conference. Details and abstract

Exercise and Sleep in People with Inflammatory Joint Diseases

The Outcome Measures in Rheumatology (OMERACT) group has identified sleep as one of the key outcomes important to people with inflammatory joint diseases (IJDs). People with various immune-mediated inflammatory diseases, including rheumatoid arthritis (RA), have reported disturbed sleep and reduced sleep duration, further adding to their disease burden. It has been well established that being physically active and taking regular exercise are important for those who have been diagnosed with an IJD. Exercise has been identified as an important part of the non-pharmacological management of poor sleep duration and in improving sleep quality. Moreover, it is known that in general, exercise improves mood state, which can also be an additional factor in improving sleep duration and sleep quality. Poor sleep has been identified as a major concern for people with RA, with disturbed sleep and fatigue known to affect up to 70% in this population.

Dr Seán McKenna and his colleagues at the School of Allied Health, University of Limerick conducted a pilot exercise trial as part of a randomised control trial (RCT) comparing an exercise programme and exercise advice for people with RA. https://link.springer.com/article/10.1007/s00296-020-04760-9

This study indicated that the overwhelming majority of those who completed the trial were surprised that exercise could help them sleep better. Sleep is an essential factor in mood, physical and cognitive performance and exercise offers a potentially attractive alternative or adjuvant treatment for those people with IJDs who have sleep issues.

This research is supported by the Irish Research Council (IRC).

Dr. Sean Mc Kenna

Irish Rheumatology Health Professionals Society

IRHPS Committee 2022/2023

Following the AGM at the Autumn meeting in Belfast September 2022 a new committee has been established.

Chairperson – Rachel Kenny RANP, Naas General Hospital

Secretary- Brid Mc Osker, Senior OT Merlin Park Hospital.

Education: Officer- Petrina Donohue, Clinical Specialist Physiotherapist in Rheumatology, Our Lady's Hospital, Navan. (Assistant Jane Brownlee Senior OT, NGH)

Treasurer: Stephanie Naramore, RANP, TUH, Dublin.

Bursary Officer: Rachel Kenny (Interim) & Aileen Woods, CNSp, MMUH

Committee Members: Maria Mc Grath, Senior PT, Peamount Health care, Martina Cooney RMDU, Harold's Cross, Angela Camon, RANP, MRHT

Please contact info@irhps.ie

@IRHPS1

HAS YOUR DEPARTMENT GOT NEWS??

Email:

info@irhps.ie

(R-L; Dr. R. Ferreria, Ms. L. Murphy, & Mr. M. Ndose

Pictured above with Ms. Louise Murphy, RANP &PhD candidate. This meeting was an excellent opportunity to network and develop research relationships.

Hypermobility in Children and Young People

Susan Ward and colleagues in Trinity College Dublin, Children's Health Ireland, University College London and University of Birmingham are studying hypermobility in children and young people.

Hypermobility describes joints that can move beyond their typical range of movement. For most, joint hypermobility causes no issues but for others it can be associated with symptoms such as pain, tiredness, and functional difficulties. Physiotherapy plays a central role in the assessment and treatment of this patient population.

A recent scoping review published by the research team identified changing characteristics from childhood to adolescence with symptoms often extending beyond the musculoskeletal system into wide-ranging clinical domains. Susan and her colleagues aim to assess clinical characteristics of children and young people with symptomatic hypermobility, using mixed methodology, integrating both quantitative and qualitative approaches to identify if clinical subgroups can be found. Following analysis, the team hopes to develop a decision-making tool to enable clinicians to target treatment. This research is supported by the National Children's Research Centre (NCRC). The NCRC has been in existence for more than 50 years and has been funded since its inception by the Children's Health Foundation Crumlin, Dublin.

IRNF
IrishRheumatologyNursingForum

The IRNF were delighted to welcome Dr Riccardo Ferreria PhD (outgoing chair of EULAR HPR) and incoming chairperson Mr. Mwidimi Ndose to the AGM in March.

Mwidimi Ndose is an associate professor, Chair of EULAR HPR and co-editor of Rheumatology and Rheumatology Advances in Practice.

National Clinical Programme:

We have contacted Gary Kileen programme manager to update the HSE website for the NCP for rheumatology. The NCP are unable to update this, this needs to be updated by the HSE.

Current Nurse Lead: Ms. Mary Gillespie

Current OT Lead: Dr. Yvonne Codd PhD

Current Physio lead: Mr. Ciaran Brennan

MSK Triage Lead: Ms. Aisling Brennan/Ms. Jenny Ashton

A number of new specialist rheumatology roles have been announced across all hospital groups as part of the "Modernised Care Pathways" for inflammatory back pain and early arthritis in a bid to achieve sustainable healthcare to move chronic health care into the community in line with Slainte Care policy. This is to improve access and improve quality within rheumatology. More information can be found at http://youtu.be/g6_kRs14NLs

Irish Rheumatology Health Professionals Society

11. World Physiotherapy Congress 2023 Submission

World Physiotherapy Congress 2023 – Submission for First Hand

I attended the World Physiotherapy Congress in Dubai this year as a first-time attendee. Arriving in Dubai, I immediately got the sense that this conference would be different. Within a few minutes of registering, I was chatting to a Physiotherapist from India, keen to show me his Instagram work account, and tell me about how he was excited about innovation and technological advances in rehabilitation. Shortly afterwards, as we headed into the exhibition hall, I found myself with a virtual reality headset on, stepping onto a virtual planet, whilst performing upper limb functional rehab. It was a unique start to a conference! The following days, I attended presentations in topics that could be considered outside of my own clinical box of paediatrics and rheumatology; planetary health, global equity, and CHAT GPT, but I quickly realised that these topics were more than relevant, challenging my own attitudes and beliefs, harnessed with new ideas and information. Other presentations that stand out include 'Gendered environment in sports', 'Pain neuroscience' and 'Beyond the biopsychosocial model'. Speakers represented all parts of the globe and brought their own cultural references to their presentations. As a paediatric physiotherapist, I attended networking sessions organised by the International Organisation of Physiotherapists in Paediatrics (IOPTP), listening to issues about access to school-based therapy in Israel, or the desire to challenge stigma towards disability in Uganda. The IOPTP were welcoming to new voices and became familiar faces at every peadiatric session. My own presentation was on day 2 of the conference, and although I was nervous, the opportunity to present our research on such a platform was fantastic. I presented the first study from my PhD, a scoping review of clinical characteristics in children and young people with symptomatic hypermobility. To my surprise, I won an outstanding platform presentation award on the final day of the conference. A huge thank you to my research supervisors, Dr Sara Dockrell, Discipline of Physiotherapy in TCD, and to Dr Emma MacDermott, in Children's Health Ireland at Crumlin, and to my co-supervisors Dr Jane Simmonds and Dr Janet Deane in University College London, and University of Birmingham. This research is funded by the National Children's Research Centre (NCRC) as a Clinical Fellowship programme. I am extremely thankful to the NCRC for their support as I near the end of this particular research journey. I now strive to complete my PhD this year, but my attendance at the World Congress will be an experience I will not forget. I encourage anyone to attend the next one, to be inspired, to be reminded of how small the world really is, and how we are all part of the bigger picture, and a global physiotherapy profession.

12. World Physiotherapy Congress 2023 Abstract

WP2023: 953

SYMPTOMATIC HYPERMOBILITY IN CHILDREN AND YOUNG PEOPLE: A SCOPING REVIEW OF CLINICAL CHARACTERISTICS USING A DEVELOPMENTAL FRAMEWORK.

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Preferred Presentation format: Platform presentation

Printed or ePoster presentation: Printed poster presentation

Primary topic: Paediatrics

2nd Topic: Musculoskeletal

3rd Topic: Rheumatology

Background: Symptomatic Hypermobility is a term used to describe the symptoms associated with joint hypermobility. Synonymous terms used include hypermobile Ehlers Danlos Syndrome and Hypermobility Spectrum Disorders. A range of musculoskeletal, gastrointestinal, cardiovascular, psychological, and urogenital characteristics are reported to be associated with Symptomatic Hypermobility. However, varied terminology across different clinical settings makes interpretation difficult in children and young people.

Purpose: The objective of this systematic scoping review was to evaluate the literature on children and young people with Symptomatic Hypermobility using a developmental framework. This review aimed to provide clarity for clinicians assessing and managing children and young people with Symptomatic Hypermobility, especially physiotherapists, who often play a key role.

Methods: The protocol was registered prospectively, according to the PRISMA-ScR guidelines. A search was conducted in EMBASE, Medline, CINAHL, Web of Science and grey literature. Full text articles with a confirmed diagnosis of Symptomatic Hypermobility, (including Hypermobility Spectrum Disorder or hypermobile Ehlers Danlos Syndrome) using internationally recognised criteria were included. Title and abstract screening and full text reviewing were conducted by two independent reviewers. Data extraction forms were piloted (two independent reviewers randomly selected n =20, approximately 12%) and were later categorised into clinical domains, and where possible, developmental stage. Children and young people were defined as; children (0-9years), adolescents (10-19years), and young adults (20-24years).

Results: Of the1619 studies screened, 163 were included in the scoping review. Studies were published from 1967-2021, 30% were narrative reviews and 24% were case reports, from a range of journals. There was only one systematic review specific to children, and it included only two studies. Musculoskeletal and cutaneous characteristics were common in all developmental stages. Cardiovascular characteristics including autonomic dysfunction, fatigue, and gastrointestinal issues were reported more frequently by adolescents. Young adult studies were scarce and included mostly case reports. Varied diagnostic criteria and assessment methods were identified. The Beighton Score was frequently used (93%), but rarely standardised (12%) and lacked consensus regarding the cut-off point of hypermobility. Gaps in evidence and specific needs for further research were identified.

Conclusion(s): This is the first systematic scoping review to identify the emerging phenotype of symptomatic hypermobility from childhood to young adulthood. This review provided evidence of the lack of guidance regarding the utilisation of standardised tools to assess hypermobility in clinical practice and research. The numerous and inconsistent diagnostic criteria and assessment methods were synthesised. Given the changing temporal pattern of symptoms, longitudinal studies and large case control studies are warranted. Such studies should have bespoke criteria and use standardised assessment protocols so that findings from different studies can be compared and data pooled for future analysis.

Implications: The identification of clear clinical characteristics could assist with the early identification of Symptomatic Hypermobility in children and young people and direct appropriate management.In future, this couldbe achieved through enhanced education of paediatric clinicians in primary and community care.

Keyword 1: Joint hypermobility

Keyword 2: symptoms

Keyword 3: clinical characteristics

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Did this work require ethics approval?: No

Institution: Trinity College, University of Dublin

Ethics committee: Faculty Of Health Sciences Ethics committee

Please state the reasons why ethics approval was not required and upload any supporting evidence: Ethics approval is not required for this Scoping Review as human participants are not involved.

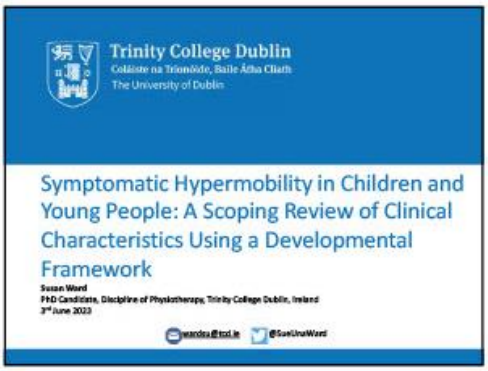
Has any of this material been/due to be published or presented at another national or international conference prior to the World Physiotherapy Congress 2023?: Yes

If yes, please provide details of where and when: Some preliminary results were submitted as an abstract to the Irish Society for Rheumatology Autumn Meeting 2021. This Scoping Review has been submitted to be considered for publication in Physiotherapy Practice and Research. This is the official journal of The Irish Society of Chartered Physiotherapists.

Consent: Yes

Consent: Yes

13. World Physiotherapy Congress 2023 Presentation



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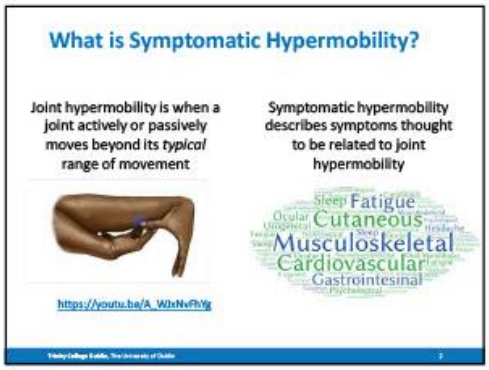

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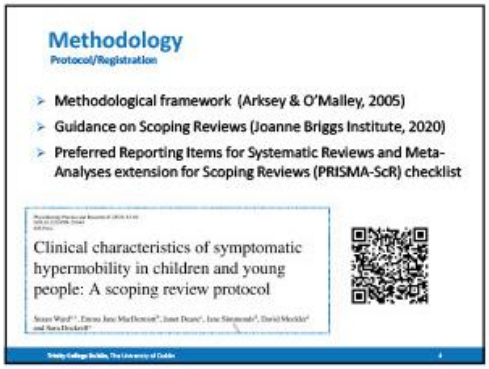

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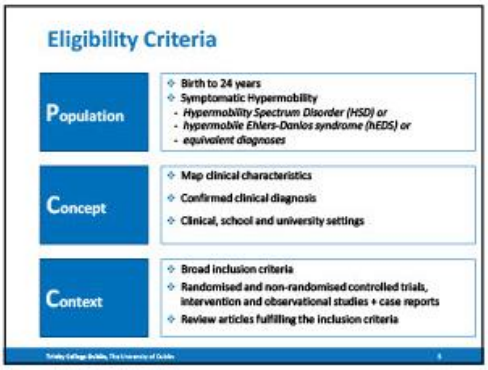

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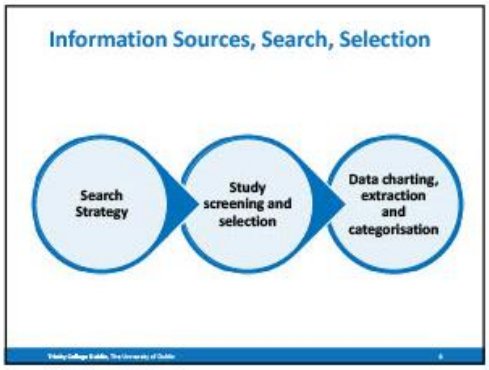

6

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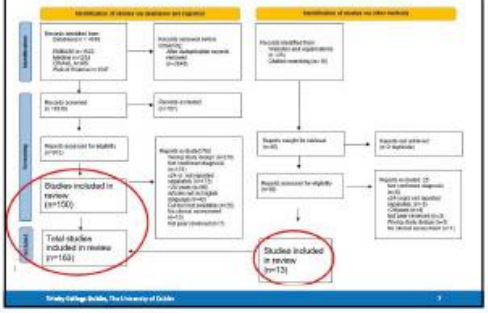
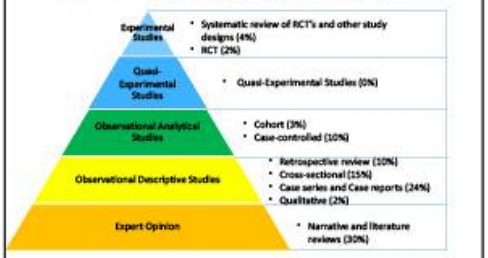
Results: PRISMA-ScR Selection of Studies



7

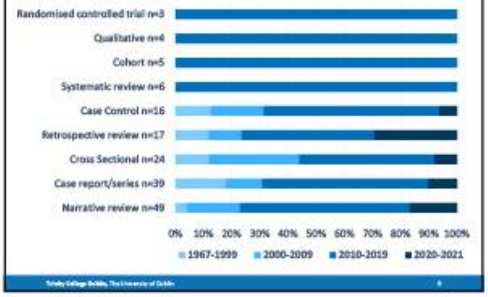
Results: Study Design by Hierarchy



8

SD1

Results: Study Design & Publication Time Frame



9

Results

CONTEXT

- **163** Studies were included
- Publication **1967-2021**
- Journal types **> 25** different clinical domains
- **26 countries**; 50% US or UK
- **N=4,444** participants included
- Settings mostly specialised clinical

MEASUREMENT APPROACHES

- **> 75** different measurement approaches
- **15** different clinical domains
- Beighton score most common (93%) but rarely standardised (12%)

10

CHILDHOOD

- 16 studies
- Musculoskeletal - pain, myalgia, arthralgia
- Cutaneous - easy bruising, soft velvety, or extensible skin
- Neurodevelopmental - gross motor delay, co-ordination difficulties

ADOLESCENCE

- 54 Studies
- Musculoskeletal - chronic pain, sprains, joint instability, dislocation
- Cutaneous- striae, delayed wound healing, soft velvety or extensible skin
- Cardiovascular - dysautonomia
- Gastrointestinal - functional GI disorders
- Urogenital - urgency
- Sleep

YOUNG ADULTHOOD

- 19 Studies
- Musculoskeletal - arthralgia, joint instability, dislocation
- Cutaneous – striae, soft velvety or extensible skin
- Gastrointestinal - functional GI disorders
- Urogenital - stress incontinence, dysuria
- Psychological- depression and anxiety

11

SD2

Discussion

- First Scoping Review in Paediatric Symptomatic Hypermobility
- Review provides greater clarity for clinicians and researchers
- Lack of consistency across studies; diagnostic criteria, measurement approaches
- Lack of standardisation; difficulty comparing studies
- Limited longitudinal studies but evidence of a changing phenotype categorised by developmental stages

12

Conclusions

- Novel synthesis of diverse literature
- Evolving phenotype was identified ⟶ need for longitudinal studies and large case control studies
- Bespoke criteria and standardised assessments
- Clinicians ⟶ recognise clinically relevant hypermobility earlier and address broad symptomology
- Consideration of care pathways beyond traditional rheumatology and genetics


Symptomatic hypermobility in children and young people: A scoping review of clinical characteristics using a developmental framework

13

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Go raibh maith agat!

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14

14. **World Physiotherapy Congress 2023 Oral Presentation Certificate**