Working Towards a Clinically Usable Allied Health Outcome Set for Paediatric and Adolescent Rheumatology Musculoskeletal Pain Conditions: A Preliminary Scoping Exercise

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ABSTRACT

Background:
Musculoskeletal conditions are very prevalent within the general population. This is also true for children and young people (CYP), with many seeking care for musculoskeletal pain. Allied Health Professionals (AHPs) are well placed to manage these patients but there is a wide variability in interventions, with no current standardisation in treatment, services, or outcomes.

Aim:
To develop a consensus-based outcome set for clinical use by AHP's working with CYP with musculoskeletal pain conditions

Method:
A staged modified nominal group technique was used. An expert panel of AHP's working in tertiary paediatric rheumatology centres in the UK identified nine domains: fatigue, muscle strength, hand function, school attendance, stamina, balance, sleep, quality of life and goal setting. Literature search and panel discussion preceded anonymous voting to select measures for each domain. Face validity of the final set was tested with a group of YP.

Results:
Consensus was achieved for six of the nine domains. Consensus could not be reached for hand function, sleep, and goal setting in part due a lack of paediatric specific measures, time taken to administer, or cost. YP agreed with the set domains but felt strongly that pain should be included.

Conclusion:
A six domain clinical outcome set has been developed for AHP's treating YP with musculoskeletal pain conditions where time, space, money, and ease of use is paramount. Future work is needed to further develop this set.

Key Points
1) Consensus-driven outcome sets for clinical use can be produced with limited funding and may be useful in guiding clinician's outcome selection in practice.
2) Young people's views into what is important in allied health professional care may vary from clinicians, and so must be included early in decisions around outcome domains and tools.
3) Further work to is needed to consider tools to capture pain symptoms as well as tools in domains that did not reach consensus and to address practical questions around outcome use and frequency in practice.

Introduction
Musculoskeletal (MSK) pain conditions are very prevalent within the general population; evidence suggests that this is also true for children and young people (CYP), with prevalence estimates ranging from 8.5-40% (Tan et al., 2018). It was once thought that MSK pain in CYP was self-limiting and without long-term impact, however, recent work has highlighted the significant impact persisting MSK pain can have on a CYP's quality of life, education, social life, sporting and leisure function and mental health (Scottish Government, 2018, Kamper and Williams, 2017, Liossi et al., 2019, Pourbordbari et al., 2019). Moreover, there can be longer term

consequences for CYP persisting with pain into adulthood, leading to increased ongoing healthcare use, long-term detriment to health and increased societal burden (Kamper et al., 2016). Significant numbers of CYP are seeking care for MSK pain, within primary, secondary, and tertiary services, and Allied Health Professionals (AHP's), with suitable paediatric experience and training, are well placed to provide interventions for these individuals. At present, however, there is significant variability in both content of and access to evidence-based interventions (Jay and Howard, 2016). Furthermore, there is little guidance on which to base provision, standardise treatment, services, or outcomes.

To address issues of health improvement and reducing health inequalities, we need robust, high quality patient outcome data to work with. Patient outcome data provides crucial clinical information to drive improvements in both the quality of, and decision-making in healthcare. The Chartered Society of Physiotherapy website states that improving the quality of healthcare in the UK is of utmost importance and that clinical outcome measures are one way of providing robust data to drive this improvement (Chartered Society of Physiotherapy, 2020). Despite this, AHP engagement in the use of outcome measurement is variable (Braun et al., 2018). Barriers to the use of outcome measures cited by professionals include time, lack of knowledge in measures to select, and lack of training in their use (Duncan and Murray, 2012).

Consensus derived outcome sets present one way to address challenges with outcome measurement and provide clinicians with an agreed set of standardised outcomes to use in a specified patient group (Williamson et al., 2012). Such sets have historically been developed for clinical trial use, aiming to align outcomes such that comparisons or meta-analyses can be more easily adopted. However, there have been sets developed that aim to guide clinical practice, as well as those aiming to cross both areas, with recognition that sets for clinical use provide their own challenges (Dodd et al., 2020).

Within paediatric and adolescent rheumatology recent advances have included developments for a minimum clinical data set for Juvenile Idiopathic Arthritis; CAPTURE (McErlane et al., 2020) and Juvenile Dermatomyositis (McCann et al., 2015). These sets are consensus driven and specify outcomes or measures which reflect current clinical practice and include both clinician-reported and patient/parent reported outcome measures.

The work reported here was initially undertaken by the British Society for Rheumatology; Paediatric and Adolescent Rheumatology (BSpaR) AHP group, who recognised the need to consider outcome measures which inform clinical practice undertaken by AHPs specifically, and to generate a set reflecting the challenges and needs of AHPs working in the UK. To our knowledge this was the first consensus driven outcome set developed for AHP use in this area. As such we aimed to scope a clinical set of outcome tools to guide AHP's working with CYP with MSK pain conditions, and as a starting point for a future research driven core outcome set. Our core objectives were:

(i) Generate a set of consensus-derived outcome measures that are widely accessible and suitable for use in a clinical setting to guide clinician use in MSK settings.
(ii) Sense-check this set within a group of service users.
(iii) Make recommendations for further work developing a robust minimal core outcome set suitable for routine patient data collection and clinical research.

Method

A multi-phased methodological approach was used, as shown in Figure 1:

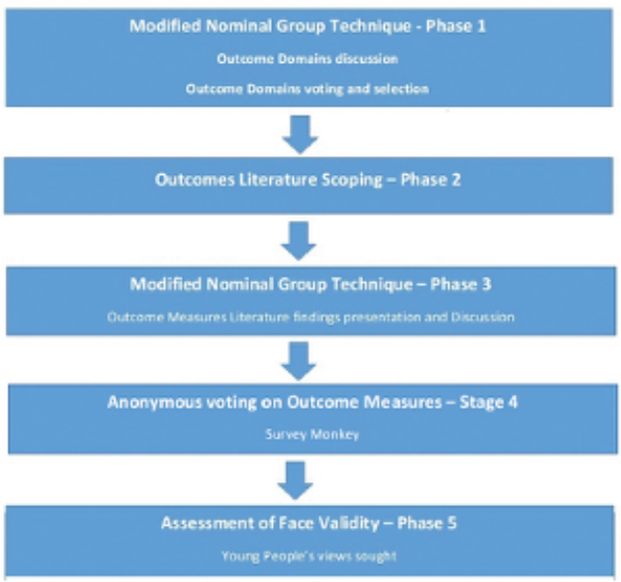

Figure 1: An overview of the multi-staged methodology used.

Phase 1: Current AHP members of BSpaR, working as senior clinicians (at least 5 years' experience specialising in paediatric rheumatology) were contacted by email and invited to participate as panel members. Fourteen clinicians, detailed in table 1, agreed to take part in this work, which was not funded and was carried out alongside usual clinical workload.

Table 1: Professions and centres of expert panel members

Profession	Physiotherapists	7
	Occupational Therapists	6
	Psychologists	1
Centre	Alder Hey Children's Hospital	1
	Birmingham Children's Hospital	2
	Bristol Royal Hospital for Children	1
	Evelina London Children's Hospital	4
	Great Ormond Street Hospital	2
	Great North Children's Hospital	1
	Royal Manchester Children's Hospital	2
	Southampton Children's Hospital	1

Initial roundtable discussion focused on the aims of the piece of work and agreeing on what domains were important for AHP's to measure. Anonymous voting on the domains listed by post-it notes was carried out by the expert AHP panel and results were used to identify the priority domains addressed in Phase 2.

Phase 2: The domains identified were shared out within the panel. An informal review of the literature was carried out for each of the outcome domains selected, to identify potential outcome measures which had been or were currently being used within paediatric musculoskeletal or rheumatology clinical settings and research studies. Electronic databases PubMed, CINAHL, Medline and web of science were systematically searched. Search terms included the domain name AND rheumatology OR musculoskeletal. Searches were limited to studies involving children, and young people, published in English, and from 2010 onwards, to limit focus to the most recent measures. Abstracts were used where possible to identify outcome measures, with full text sought if this was not possible. Additionally, any measures currently used by panel members or colleagues in clinical practice with the defined population were included.

Phase 3: A meeting of the panel was convened. Each domain and associated literature findings were presented to the panel with discussion facilitated by chairperson to focus on the advantages and disadvantages of each

measure. Discussion was guided by the principles for outcome tool selection suggested by Prinsen et al (Prinsen et al., 2016) with particular focus on:

- Validity within a paediatric and adolescent population
- Use in a variety of clinical settings
- Cost to obtain
- Ease of use with minimal training

Phase 4: Following the face-to-face panel meeting, a survey, using survey monkey (www.surveymonkey.com), was sent to each of the panel members by email, asking the participants to choose the strongest measure for each domain, measured against the list above, or none, if none of the measures were felt to fulfil the criteria. Consensus was pre-determined as agreement between experts, participating in the online survey, of 60% or more (Fink et al., 1984).

Phase 5: The consensus findings were sense checked by members of the national youth advisory panel in adolescent and young adult rheumatology – Your Rheum (http://yourrheum.org; (McDonagh et al., 2019)). Meetings with young people were conducted without presence of clinicians to reduce potential influence by health professionals and to optimise the willingness of the young people to input authentically to the discussion. The meeting involved three young people aged 12, 18 and 24. An arts-based approach was selected for this event in order to generate thoughts, ideas and discussion about the topic prior to the use of open-ended questions about the COS. Art-based approaches are often used in eliciting YP views around health-related issues and are frequently used in conjunction with interviews or discussion groups (Coad, 2007). Table 2 shows the methods used for the face validity testing.

Table 2: Arts-based face validity testing method

Step One	Young people (YP) were given a size A0 "life course map" and a variety of craft materials: pens, pencils, magazines, scissors, glue etc. Instruction was given to consider broadly the outcomes, their indicators of health and well-being and key events throughout childhood/adolescence and young adulthood that were important and had meaning to them.
Step Two	YP were asked to record outcomes, in any medium available, at the point on the life course map where they felt they fitted best. It was impressed upon the YP that there was no right or wrong in this situation and that they were free to record and express anything that they felt was of importance within this topic area.
Step Three	Following this the outcomes identified by the expert AHP group were introduced to the YP. They were invited to comment openly on the domains, specifically: • Did they agree or disagree with the items included? • Was there anything that had not been included that should have? YP feedback was documented during the discussion and used to inform the final COS.

Results

Phase 1 findings: Nine domains were agreed on. These were fatigue, sleep, school function, achievement of goals, muscle strength, balance, stamina, hand function and quality of life.

Phase 2 findings: Literature scoping identified 39 potential measures across the 9 domains (See appendix 1).

Phase 3 and 4 findings: The response rate for the expert panel survey was 83% (n=10 of 12). Consensus was derived for 6 of the 9 domains considered, with no consensus for sleep, achievement of goals or hand function. Table 3 shows the agreed core outcome set. Fatigue, muscle strength and stamina produced a tied result and so both measures have been included at this stage.

Table 3: List of agreed measures for core outcome set

Domain	Outcome Measure
Fatigue	Fatigue Visual Analogue Scale (Crawford et al., 2011), Paediatric Quality of Life Multidimensional Fatigue Scale (Varni et al., 2004)
School function	Attendance rate (Weitzman, 1986)
Muscle Strength	Kendal scale (Kendall et al., 1993), Oxford scale (Compston, 1942)
Balance	Standardised Single Leg Stance (Condon and Cremin, 2014)
Stamina	Timed step test (Balfour-Lynn et al., 1998), Two minute walk test (2MWT) (Bohannon et al., 2014)
Quality of life	Paediatric Quality of Life Inventory Generic Core Measure (Peds QL) (Varni et al., 1999)

Phase 5 findings: Discussion with YP found agreement with all domains included in the COS, and they felt that they were all of importance to their daily lives and functioning. However, they felt very strongly that pain was a domain that should be included due to the large impact it has on their day-to-day life.

Discussion

A preliminary core outcome set for clinical use by allied health professionals working with children and young people with musculoskeletal pain conditions has been developed, comprising of six consensus agreed outcome measures for fatigue, school function, muscle strength, stamina, balance, and quality of life. Consensus could not be reached for measures for sleep, goal setting and hand function. Possible reasons for this include identified tools not having suitable psychometric properties, being expensive to obtain, being time consuming to use in clinical practice and lacking appropriate validity testing.

Core outcome sets are an accepted way of agreeing a standardised minimum set of outcome measures (Williamson et al, 2012). Interest in the development of such sets has resulted in a series of published methodological guidance The COS-STAD checklist for outcome selection, along with guidance on reporting, protocol publishing and outcome tool selection (Kirkham et al., 2017, Kirkham et al., 2016, Kirkham et al., 2019, Prinsen et al., 2016) is available to guide researchers. The methodology used here has several strengths, including a clear setting, population, condition, and interventions, allowing clinicians to judge the suitability of the set for their use. However, there are several limitations. Unlike traditional COS development, both domain selection and outcome tool selection were encompassed within the one multi-phased study. Full COS development places more emphasis on psychometric evaluation rather than feasibility of use, setting a minimum standard for inclusion of an outcome measure (Prinsen et al., 2016). Barriers to outcome measure use for AHPs highlight lack of time as a key factor in uptake (Duncan and Murray, 2012). Therefore, we made the decision at this early stage to prioritise feasibility over psychometric properties, agreeing validation in a paediatric population, as minimum criterion. A further limitation was in stakeholder inclusion. The stakeholder group included representation from eight centres and three professional groups but lacked input from either CYP or parents. Patient involvement is noted as important in COS development (Kirkham et al., 2017, Williamson et al., 2012). We were able to seek the opinions of CYP through the research involvement group Your Rheum (www.yourrheum.org). Due to resources this was limited to a retrospective sense checking activity. This was attended by three YP, aged between twelve and twenty-four, with no inclusion of parents or younger children. As this activity was conducted through the Patient and Public Involvement (PPI) framework and organised by Your Rheum, this limited the research groups' input into the number of participants involved. We recognise the limitations of this small sample size, and the associated impact of limiting the generalisability of findings to a wider population. Additionally, the narrow age range restricts the insight gained to the adolescent population, with additional work needed to consider use of the set in a younger population.

As the first allied health clinical core outcome set in paediatric musculoskeletal conditions it is not possible to directly compare our findings to other studies. Other, comparable work in progress includes an arthritis symptom tracker for teens with JIA. This project has sought the opinions of young people with JIA on which items and symptoms they felt were most important to monitor. YP identified energy levels, medication side effects, activity levels and sleep function as important to track the course of their disease (Versus Arthritis, 2020). These map well to the outcomes of fatigue, stamina and strength, and sleep identified in this study, with medication side effects not directly within allied health professionals' scope of practice. However, there were areas of disagreement between this project and our core outcome set.

When considering domain inclusion, pain was excluded by the professional panel in the early stages. This was because the panel prioritised functional outcomes, as it was felt that focus on controlling pain may lead to prioritisation of pain symptom management over functional improvement. This has been found to be unhelpful in longer term instances of pain, and can result in dysfunctional beliefs by young people and their parents around pain (Caes et al., 2012). Additionally, it was felt that the known complexity of pain assessment, as well as the complex interaction of pain and developmental stage would pose a challenge outside the scope of this work. The YP involved in our sense-checking strongly disagreed with the panel, which aligns with findings from the Versus Arthritis app project, where pain was a symptom prioritised by YP to be tracked (Versus Arthritis, 2020). The YP we consulted were clear that pain was central to their function, was needed for interpretation of other results and that it should be personalised to the young person and measured specifying context.

Parents and YP often differ in their treatment priorities to professionals (Sinha et al., 2012, Sherratt et al., 2020), as has been found to be the case in this work. Whilst the panels concern around pain behaviour may be relevant, ensuring that clinical outcomes used are important and meaningful to YP is crucial in ensuring a shared focus for treatment. Failure to capture outcomes deemed relevant to YP may limit the use of clinical decision making, and impair engagement and trust between YP and their clinicians (Sherratt et al., 2020) Whilst the PedsQL generic core measure, recommended as the quality-of-life tool, does include items that related to pain symptoms, pain should be looked at as a further outcome domain. Future work is needed to agree on an appropriate pain outcome tool for use in practice and this work should prioritise engagement of young people early in the research process.

This preliminary piece of work is a valuable starting point for aiding clinical selection of outcome measures. Along with the addition of pain, future work should explore further the measures with tied consensus to allow the recommendation of one measure per domain (Prinsen et al., 2016). Additionally, consideration should be given to those domain tools that did not reach the pre-agreed consensus- sleep, hand function and achievement of goals. Psychological outcomes should also be considered, as the interplay between physical and psychological health and wellbeing has long been recognised. Psychologists have an important role to play in the management of paediatric MSK conditions (Davis et al., 2017), although access is variable nationally (British Society for Rheumatology, 2019, Hawley et al., 2018).

Advice on timing and frequency of outcome collection is also important to consider. AHP models of care vary from blocks of more intense treatment for some patients, to regular review throughout childhood and adolescence to account for developmental changes and to provide developmentally appropriate care. ARMA (Arthritis and Musculoskeletal Alliance) standards for CYP with JIA for example, suggest a minimum of an MDT review annually (Arthritis and Musculoskeletal Alliance, 2010), however this is often at odds with current NHS policies, with patients with long term conditions being discharged from treatment. AHP care models for paediatric MSK conditions vary nationally and will impact agreement on the timing and frequency of COS collection.

Conclusion

This scoping work represents, to the best of our knowledge, the first allied health specific outcome set led by allied health professionals for their clinical use in Paediatric Rheumatology settings. We recommend an initial consensus derived set of outcome measures to assist allied health professionals in the selection of tools for assessing outcomes in young people with musculoskeletal pain conditions. We recognise the limitations to this work and identify key recommendations points for future projects to further develop this core outcome set.

This is an important collaborative step by the BSpaR AHP group to challenge variation in paediatric MSK service provision and work towards driving improvements in care and informing decision-making for this population.

Ethical Approval:

Ethical approval was not sought for this study: attendance by AHP's at the expert panel meeting was considered as consenting to participate in the consensus process. Consent by CYP at the face validity testing was sought by YOURRHEUM personnel and was done within the PPI framework. As such the young people were not research participants and therefore ethics approval was not required (National Research Ethics Service and INVOLVE, 2009).

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Appendix 1: All outcome measures identified and considered by expert panel

Domain	Measures considered	Reference
Fatigue	Fatigue Visual Analogue Scale (Fatigue VAS)	Crawford BK, Piault EC, Lai C, Bennett RM. Assessing fibromyalgia-related fatigue: content validity and psychometric performance of the Fatigue Visual Analog Scale in adult patients with fibromyalgia. Clinical and experimental rheumatology. 2011;29(6 Suppl 69):S34-43.
	Paediatric Quality of Life Inventory (Peds QL) Multidimensional Fatigue Scale	Varni JW, Burwinkle TM, Szer IS. The PedsQL™ multidimensional fatigue scale in pediatric rheumatology: Reliability and validity. Journal of Rheumatology. 2004;31(12):2494-500.
	Computerised Adaptive Test for Fatigue in Rheumatoid Arthritis (CAT Fatigue RA)	Nikolaus S, Bode C, Taal E, Vonkeman HE, Glas CAW, van de Laar MAFJ. Construct Validation of a Multidimensional Computerized Adaptive Test for Fatigue in Rheumatoid Arthritis. PLOS ONE. 2015;10(12):e0145008-e.
	Bristol Rheumatoid Arthritis Fatigue Scales (BRAF)	Nicklin J, Cramp F, Kirwan J, Greenwood R, Urban M, Hewlett S. Measuring fatigue in rheumatoid arthritis: A cross-sectional study to evaluate the Bristol Rheumatoid Arthritis Fatigue Multi-Dimensional questionnaire, visual analog scales, and numerical rating scales. Arthritis Care & Research. 2010;62(11):1559-68.
Sleep	Multidimentional Fatigue Symptom Inventory-Short Form (MFSI-SF)	Stein KD, Jacobsen P, Thors C Further validation of the multidimensional fatigue symptom inventory-short form. Journal of pain and symptom management. 2004;27(1):14-23 .
	Children's Sleep Habits Questionnaire (CSHQ)	Owens JA, Spirito A, McGuinn M. The Children's Sleep Habits Questionnaire (CSHQ): Psychometric Properties of A Survey Instrument for School-Aged Children. Sleep. 2000;23(8):1-9.
	Bedtime problems, Excessive daytime sleepiness, Awakenings during the night, Regularity and duration of sleep, Snoring (BEARS)	Owens JA, Dalzell V. Use of the 'BEARS' sleep screening tool in a pediatric residents' continuity clinic: A pilot study. Sleep Medicine. 2005.
School Function	Attendance Rate	Weitzman M. School absence rates as outcome measures in studies of children with chronic illness. Journal of Chronic Diseases. 1986;39(10):799-808.
	School Setting Interview (SSI)	Hemmingsson H, Borell L. The Development of an Assessment of Adjustment Needs in the School Setting for Use with Physically Disabled Students. Scandinavian Journal of Occupational Therapy. 1996;3(4):156-62.

	School Function Assessment (SFA)	Coster WJ, Mancini MC, Ludlow LH. Factor Structure of the School Function Assessment. Educational and Psychological Measurement. 1999;59(4):665-77.
Achievement of Goals	Goal Attainment Scale (GAS)	Steenbeek D, Ketelaar M, Galama K, Gorter. JW. Goal attainment scaling in paediatric rehabilitation: a critical review of the literature. Developmental Medicine & Child Neurology. 2007;49(7):550-556.
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