“Sticking to it”

A Scoping Review of Adherence to Exercise Therapy Interventions in Children and Adolescents with Musculoskeletal Conditions.

by

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Abstract

Background: Exercise therapy is a core component of treatment for children and adolescents with musculoskeletal conditions. As exercise therapy effectiveness hinges on adherence, improving exercise therapy adherence is crucial for recovery and reducing long-term consequences.

Objective: This scoping review consolidates the breadth of knowledge about exercise therapy adherence barriers, facilitators, and boosting strategies for youth with musculoskeletal conditions. In doing so, this review will inform clinical practice and future research.

Methods: This review was guided by Arksey and O'Malley's 5-stage framework and the PRISMA Extension for Scoping Reviews. Six electronic databases were searched using predetermined search terms and Medical Subject Headings. English studies with original data featuring an adherence barrier, facilitator, or boosting strategy and youth (≤19 years) with musculoskeletal conditions treated with exercise therapy were included. Two authors independently conducted title/abstract and full-text reviews. Study quality was assessed using the Mixed Methods Appraisal Tool. Descriptive consolidation and thematic analysis were completed using the Capability, Opportunity, Motivation and Behaviour (COM-B) framework.

Results: Of 4,930 potentially relevant records, 34 studies representing 1,563 participants (65% female, 2-19 years of age) with 11 different musculoskeletal conditions and multiple exercise therapy interventions were included. Across studies, adherence concepts were poorly reported with adherence rates ranging from 15%-99% of prescribed exercises. Time constraints, physical environment (e.g., location), and negative exercise experiences were commonly identified adherence barriers, while social support and positive exercise experiences were frequently

identified facilitators. Reinforcement, exercise program modification, and education were commonly used adherence boosting strategies, despite being infrequently reported as barriers or facilitators. Exercise experience (positive/negative), time, and environment (physical and social) emerged as important themes related to exercise therapy adherence for youth with musculoskeletal conditions.

Conclusion: Despite poor reporting of adherence concepts, a diversity of barriers and facilitators to exercise therapy for youth with musculoskeletal conditions exist. Existing strategies to boost adherence are not consistent with identified barriers or facilitators. Making exercise enjoyable, social, and convenient may be important to maximizing adherence in this population. Efforts to match adherence boosting strategies to an individual's needs should be considered and should respect the key themes.

Preface

This thesis is an original work by Christopher James Holt. No part of this thesis has been previously published.

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1. Introduction

Exercise therapy is a vital part of treatment for children and adolescents with musculoskeletal conditions. Unfortunately, many youth (broadly defined as children and adolescents ≤19 years of age) do not complete the exercise therapy programs as prescribed. This can delay recovery, contribute to ongoing pain, and put youth at risk for serious medical conditions later in life. For youth, the challenges associated with adhering to an exercise program are complex. Understanding how best to help youth with musculoskeletal conditions "stick to" their prescribed exercise programs is essential to reducing the long-term consequences of these conditions and encouraging lifelong health and mobility. To further our understanding on this topic, a comprehensive review of the evidence-base is required to consolidate existing knowledge and highlight directions of future study.

1.1 The Burden of Youth Musculoskeletal Conditions

The world wide burden of musculoskeletal conditions (i.e., diseases and injuries) is well known,[1–3] affecting people of all ages. The prevalence of musculoskeletal pain is estimated as high as 27-36% in adolescents.[4] Further, a staggering 29% (95% Confidence Interval (CI), 27.1, 31.8)[5] of Canadian youth aged 12-15 years and 40% (95% CI 38.4, 42.1)[6] of youth aged 14-19 years seek medical attention for an musculoskeletal injury annually, with resulting healthcare costs estimated at $46.7 million (CAD) annually.[7] The link between childhood musculoskeletal injury and future health conditions is also well documented.[3,8] For example, it is estimated that youth who suffer a significant knee injury are 3.75 times (95% CI 1.2, 11.3) more likely to be overweight/obese 3-10 years post injury when compared to uninjured youth.[8,9] This elevated risk is particularly alarming given that obesity is associated with high rates of morbidity (i.e., osteoarthritis[8] and cardiovascular disease[10]) and all-cause mortality,[11,12] and underlines the long-term importance of appropriate care for youth with musculoskeletal conditions.

1.2 Exercise Therapy in the Management of Youth Musculoskeletal Conditions

While best practice recommendations for musculoskeletal conditions vary across condition, location, and injury type, evidence-based treatment guidelines commonly include patient-centered care, education, and exercise therapy.[13–15] "Exercise therapy" is defined as a regimen or plan of physical activities designed and prescribed in a precise dose to address specific therapeutic goals such as restoring musculoskeletal function or reducing pain caused by disease or injury.[16] Distinct from physical activity, it can include strengthening, aerobic, and/or motor

control exercises, and can be administered in supervised (e.g., in clinic) and/or non-supervised environments (e.g., home exercise program). Many studies have shown exercise therapy as a feasible and effective option for youth with musculoskeletal conditions, including systematic reviews of youth with juvenile idiopathic arthritis[17] and low back pain,[18] and randomized control trials of youth with patellofemoral pain and idiopathic scoliosis.[19,20] These studies have shown exercise therapy can improve symptoms, function, and quality in life, in both the short and long term for youth with musculoskeletal conditions.[17–20]

There are many physiological factors unique to youth that impact prescription of exercise therapy when compared to adults.[21] To ensure exercise therapy is safe, physical characteristics such as physiological maturation, skeletal maturity, and movement pattern development must be considered.[22] In addition, cognitive factors such as differences in maturity, motivation, communication skill, and the ability to comprehend/follow instructions may also impact safety.[21,23] With this in mind, several safe exercise training guidelines for youth have been recently published,[24,25] including an international consensus on youth resistance training (2014).[26]

In addition to physiology, unique social contexts must also be considered when prescribing exercise therapy to youth.[21] For example, daily routines are often busy and non-flexible (e.g., school and recreation schedules), influencing treatment session attendance and participation in home exercise therapy programs. Youth are less autonomous than adults and often dependent on guardians for scheduling, transportation, and environment, whether by choice or necessity.[27] Additionally, peer groups are known to have a much larger influence on adolescents' behaviour than on adults', which may impact the desire to complete treatment and their resiliency during rehabilitation.[21,28] All of these factors (i.e., physiological and social) must be considered when prescribing exercise therapy programs to youth in order to maximize safety, effectiveness, and participation.

1.3 Adherence terminology

In the context of prescribed exercise therapy, "participation" is commonly referred to as "compliance" or "adherence". While often used interchangeably, these terms do differ when describing the degree to which someone engages in exercise therapy. "Compliance" refers to the act of an individual conforming to recommendations for a prescribed dosage of exercise therapy (e.g., timing, frequency, load).[29] Compliance is most commonly used in research settings when a consistent intervention is desired across participants to measure the efficacy of an exercise

therapy intervention. In contrast, “adherence” describes the collaborative planning process that takes place between a clinician and patient to effect long-term, sustainable behaviour change.[30] Adherence emphasizes the importance of patient autonomy and empowerment when evaluating treatment adoption, and is often used in studies of intervention effectiveness, rather than efficacy.[29–31] “Adherence” is therefore often the preferred terminology when discussing treatment participation in the context of how behaviour change can be facilitated in “real world” settings.[29,31] Despite calls to consider “compliance” and “adherence” in this way, adherence terminology remains poorly defined and inconsistently used across studies.[32]

1.4 The Relationship Between Exercise Therapy Adherence and Clinical Outcomes

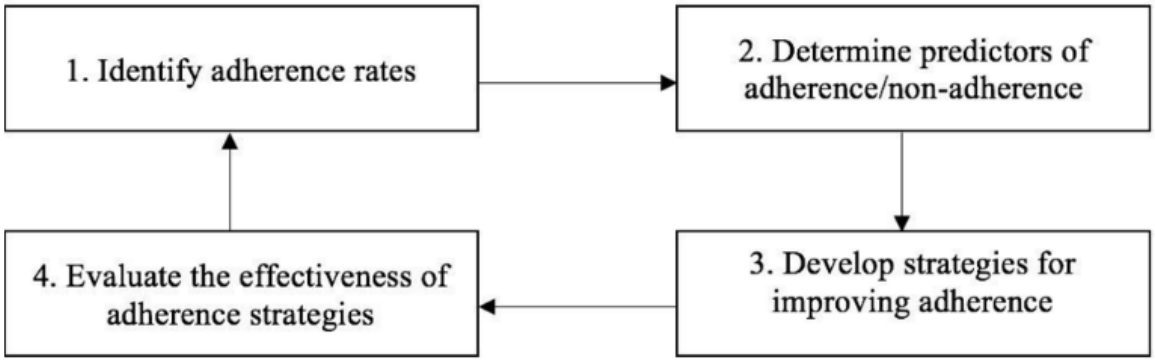
Predictably, many studies have reported high adherence to exercise therapy is associated with improved clinical outcomes.[20,33–35] For example, in youth (age 15-19 years) with patellofemoral pain, a >70% treatment adherence (Percentage of Prescribed, POP) was associated with a 4-fold greater odds (OR 4.05; 95% CI 1.42, 11.55) of 12-month recovery compared to ≤70% adherence.[20] Similarly, in youth (age 10-14 years) with adolescent idiopathic scoliosis, exercise therapy compliance has been shown to be strongly associated with spinal curve improvement.[35] Despite this relationship, there remains no gold standard for defining “acceptable” adherence, or the minimum dosage required to achieve clinical effect.[32] Bailey et al[32] reported that amongst 71 studies investigating exercise therapy adherence for treating adult musculoskeletal pain, the minimum satisfactory level of adherence varied widely, and in many cases appeared to be arbitrary, lacking justification.[32]

Given the association between adherence and clinical outcomes, it is concerning that adherence to exercise therapy in both adult and youth populations is consistently reported as low. For example, in one study of patients attending outpatient physiotherapy for a variety of musculoskeletal conditions, only 35% were fully adherent with their exercise program.[36] Specific to youth, one study reported 40% of participants aged 12-16 years with patellofemoral pain attended less than 40% of prescribed exercise sessions, and no participants met the goal of attending 80% of sessions.[37] Consistently poor adherence rates have highlighted the need to develop improved adherence boosting interventions for this population and to understand what impacts exercise therapy adherence in youth.

1.5 Improving Exercise Therapy Adherence

In 2018, Owoeye et al[38] proposed a theoretical model for improving adherence to exercise therapy. This model outlines four distinct steps required to improve adherence (see Figure 1): identifying adherence rates, determining predictors of adherence/non-adherence, developing strategies for improving adherence, and evaluating the effectiveness of adherence strategies. This model clearly indicates the importance of identifying predictors of adherent behaviour and valid and reliable measurement outcomes prior to, or alongside, the development of boosting strategies.

FIGURE 1. Owoeye's model for improving exercise adherence[38]



1.5.1 Identifying Adherence Rates

Considerable variability exists in how adherence to exercise therapy has been operationalized.[32] Possible explanations for this variability include the abstract nature of adherence, the multidimensional nature of exercise therapy, and the diversity of settings in which exercise therapy can be completed. Common adherence outcomes (i.e. quantifiable variables) include: total number of exercise sessions attended, frequency of exercise sessions completed, time spent exercising, or the level of exercise exertion or intensity.[32] However, regardless of outcome used, adherence to exercise therapy is most commonly reported as the Percentage of Prescribed (POP) exercises that are completed. In some fields, efforts have been made to standardize how adherence is operationalized to enable data amalgamation and comparisons between investigations.[38] For instance, the concepts of exercise utilization frequency (cumulative sum of exercise sessions completed) and exercise utilization fidelity (components completed of total possible per session) have been proposed in the sport injury prevention field. However, the uptake

of these concepts has been slow, and applicability of these recommendations across fields remains unclear.[38]

There is also a lack of consensus about methods or instruments to collect adherence to exercise therapy data. Three recent systematic reviews identified over fifty unique measurement tools.[39–41] The majority of these tools were "self invented",[41] had unknown measurement properties,[40] and were rated as "poor" methodological quality (Consensus-based Standards for the Selection of health Measurement Instruments (COSMIN) checklist).[39] In adults with chronic musculoskeletal conditions, the most common approach to collecting adherence is a home diary.[41] However, self-report approaches (e.g., home diaries, exercise logs) are prone to recall and subsequent measurement bias, and reportedly overestimate adherence by as much as 25%.[42] Regardless of exercise setting, there is a clear need for well-developed universal approaches to capture adherence to exercise therapy interventions across ages.[43]

1.5.2 Determining predictors of adherence/non-adherence

According to the World Health Organization, ''the ability of patients to follow treatment plans in an optimal manner is frequently compromised by more than one barrier, usually related to different aspects of the problem.''[30,44] Therefore, identifying the possible barriers and facilitators associated with exercise therapy adherence is a key preliminary step to developing interventions aimed at improving adherence. Several systematic reviews have identified the common barriers and facilitators to exercise therapy adherence in adults.[44,45] Essery et al[44] reported strong evidence for barriers such as number of exercises, stress levels, and forgetfulness, as well as facilitators such as self-motivation, intention to adhere, and social support.[44] Jack et al[45] found barriers to outpatient exercise therapy following musculoskeletal injury included anxiety/depression, pain during exercise, and reduced self-efficacy.[45] To date, no reviews have examined barriers and facilitators to exercise therapy adherence in youth with musculoskeletal conditions.

1.5.3 Developing strategies for improving adherence

Beyond reducing barriers and promoting facilitators, specific behaviour modifying interventions have been hypothesized to improve exercise therapy adherence. For example, formal goal setting and motivational techniques have been employed to improve adherence to exercise therapy in adults with musculoskeletal conditions.[46–48] Gamification, live feedback, and incentive programs have also been hypothesized to improve adherence through psychosocial pathways.[49–51] Unfortunately, not all strategies have been adequately evaluated, and of those that have, there is

a lack of general agreement between clinicians and patients as to which strategies are preferable or effective.[47] This suggests that the development and evaluation of acceptable adherence boosting strategies is another area in need of high quality evidence, specifically as it applies to youth populations. From a practical perspective, it is important to consider that there is considerable variability amongst patients and that there is no specific profile associated with being a “non-adherer”. Therefore, it is likely that the most prudent approach to improving adherence to exercise therapy is one that employs a variety of individualized strategies and techniques.[52,53]

1.6 Adherence and behaviour change theory

As adhering to exercise therapy involves the adoption of a new behaviours, behaviour change theory can provide a useful conceptual basis for understanding adherence. Further, behaviour change theories can be used to guide the choice of appropriate behavior change techniques to improve implementation of an exercise therapy program. Michie et al[54] suggested one such theory (the COM-B Framework, 2011) for categorizing the wide variety of factors (e.g., barriers, facilitators, and strategies affecting exercise therapy adherence) into the required elements for adopting a specified behaviour.[54] This framework categorizes these factors into someone’s Capability to adopt (both physical and psychological capacity to engage in activity), Opportunity to adopt (external factors including both their physical and social environment), and Motivation to adopt (brain processes including both automatic and reflective reaction to motivating factors) a specific behaviour.[54] According to Michie et al[54], these elements all interact to generate a specific behaviour (e.g., adherence), and consideration of these elements can help with successful intervention planning, and in identifying areas in need of further development.[54] To assist with implementation and intervention planning when applying the COM-B Framework, the Theoretical Domains Framework has also been developed for classifying determinants of behaviour and providing practical guidance.[55]

2 Knowledge Gaps Associated with Adherence to Exercise Therapy in Youth with Musculoskeletal Conditions

Despite the association between exercise therapy adherence and clinical outcomes for youth with musculoskeletal conditions, exercise therapy adherence amongst this population remains poor,[36,37] and a common source of clinical frustration. Proposed steps for improving adherence include determining the predictors of adherent behaviour (i.e., barriers and facilitators).[38] As predictors of exercise therapy adherence in youth with musculoskeletal conditions remain unclear, a wide variety of adherence boosting interventions have been proposed. However, to date no consensus exists on which interventions are most effective. These gaps in knowledge are significant hurdles to optimizing adherence to youth exercise therapy, improving clinical outcomes, and thereby decreasing the long-term impact and global burden of youth musculoskeletal conditions. A comprehensive review of the evidence-base is required to address the gaps in knowledge related to exercise therapy adherence in youth with musculoskeletal conditions, to consolidate existing knowledge, and highlight directions of future study.

3 Objectives

The *primary objective* of this scoping review is to consolidate primary peer-reviewed evidence regarding exercise therapy adherence barriers, facilitators and boosting strategies (interventions) in youth with musculoskeletal conditions. *Secondary objectives* include mapping out relationships between known barriers and facilitators to adherence and current strategies to improve adherence, and identifying gaps in the existing evidence-base.

4 Methods

4.1 Framework, protocol, and registration

This scoping review follows the 5-step methodological framework proposed by Arksey and O'Malley[56] with consideration of subsequent recommendations by Levac et al[57] and the Joanna Briggs Institute.[58] The members of the study team were selected to ensure expertise in quantitative methodology (JW, DG, CM), behaviour change theory (CM), clinical prescription of exercise therapy (CH, LT, CL, DG, JW), and pediatric rehabilitation (CH, JW). Reporting follows the PRISMA Extension for Scoping Reviews (PRISMA-ScR)[59] which recommends subtle changes to language and organization from previous versions[60] to help reflect the diverse evidence associated with scoping methodology (Appendix A).[59] For example, "information sources" refers to where records are compiled from (e.g., bibliographic databases, social media platforms, web sites), while "sources of evidence" is recommended as a more inclusive and heterogeneous term for eligible records (e.g., quantitative and/or qualitative research, expert opinion, and policy documents).[59] In addition, we adopted the recommended term "data charting" in lieu of "data extraction" to better reflect the "sifting, charting, and sorting"[56] of diverse material commonly retrieved in scoping reviews.[56–58] As there was no database for scoping review protocol registration in existence at the initiation of this project, the protocol was not registered a priori.

4.2 Information sources and search

Six online databases were selected based on relevance to the field of exercise therapy and included the Medical Literature Analysis and Retrieval System Online (MEDLINE), the Cumulative Index of Nursing and Allied Health Literature (CINAHL), SPORTDiscuss, Scopus, the Physiotherapy Evidence Database (PEDro), and ProQuest. Databases were searched using a combination of Medical Subject Headings (MeSH) and keywords selected based on consultation with a librarian scientist and content experts from amongst the study team (JW, DG, CM). A copy of the complete search strategy (including all MeSH headings and subject keywords) is available in Appendix B. Due to the broad nature of "musculoskeletal injury/condition", the initial search did not include a musculoskeletal concept to ensure no record was missed due to misclassification of the condition. Instead, the study team agreed to inspect search results for condition by hand during title/abstract screening. Searches were organized using the reference management software EndNote X8.2. Internal (within database) and external (between databases) duplicates were removed from results and saved for future reference prior to screening.

4.3 Sources of evidence (study selection)

Sources of evidence were included if they discussed an outcome or construct of adherence to an exercise therapy intervention in adolescents or children (ages ≤19 years) with a musculoskeletal condition (i.e., disease or injury). Studies were excluded if they were not written in English or involved animal models or cadavers. Additional inclusion criteria included primary research with original data (i.e., peer-reviewed publications and theses) inclusive of experimental, quasi-experimental, observational, and qualitative designs. Conference proceedings and abstracts were excluded due to the unlikelihood of adherence concepts being discussed in detail. Given the objective to map out peer-reviewed primary (i.e., original data) research, editorials, commentaries, opinion-based papers, and review articles (systematic and narrative) were excluded. The reference lists of all review articles were hand searched for additional relevant records. Inclusion criteria regarding "adherence" (i.e., must identify an adherence barrier, an adherence facilitator, or an adherence boosting strategy) was only applied during full text review stage to ensure no sources of evidence were excluded if adherence was not featured in the abstract. Complete inclusion/exclusion criteria (with justification) are listed in Appendix C.

Prior to title/abstract screening, all reviewers (CH, CM, LT, CL, DG) independently screened a random sample of 120 titles and abstracts to assess the applicability of the exclusion criteria and determine the inter-rater agreement with the senior author (JW). All reviewers reached "moderate" to "almost perfect" agreement with the senior author (agreement ranging from 84% to 97%, Cohen's *Kappa* ranging from 0.45 to 0.84). Following this step, discrepancies were discussed between reviewers, criteria were iteratively adjusted, and the exercise was repeated until confidence in the application of the exclusion criteria was achieved.

After accounting for duplication, the titles and corresponding abstracts of all returned records were independently screened by two authors blinded to record author(s) and journal title using a Microsoft Excel workbook designed specifically for screening.[61] Discrepancies between reviewers were resolved through discussion, and, if needed, through consultation with the senior author (JW). Following removal of records excluded at title/abstract screening stage, full-text copies of remaining studies were then located using online records, interlibrary loan, or direct contact with the study author (CH). Full text was then reviewed, and inclusion criteria applied by two independent raters, with discrepancies resolved as above.

4.4 Data charting

Data charting was completed by the lead author (CH) using a custom designed (CH, LT, JW) Microsoft Excel workbook (Office 365 MSO, 2018). Prior to data charting, the tool was independently piloted and refined by the lead (CH) and senior author (JW) on a purposive sample of included studies of various study designs. Items for data charting were selected through study team discussion (CH, LT, JW) using the Joanna Brigg's Institute data charting recommendations.[58]

Data items charted from each source included: author, year, title, study design, study objective, sample characteristics (i.e., size, age, sex, and condition), exercise setting (i.e., hospital, community centre, clinic, school, or home), exercise supervision (i.e., full, partial, or no supervision), exercise session frequency (i.e., days/week prescribed), exercise session length (i.e., minutes/session prescribed), exercise program length (i.e., weeks prescribed), adherence targets or goals (i.e., percentage of prescribed), adherence rates (i.e., percentage of prescribed), adherence outcomes, adherence outcome measures, barrier(s) or facilitator(s) to exercise therapy adherence, and strategies or interventions used to improve adherence. For this review, strategies or interventions used to improve adherence were defined as a specific action discussed and implemented by the researcher a priori with the intent of improving adherence. The efficacy of these interventions to improve adherence was not collected in this review due to the wide variety of study quality and interventions identified, and because efficacy of these strategies was not examined in most studies. The data-charting tool, complete with all data items from all sources, is included in Appendix D.

4.5 Critical appraisal of individual sources of evidence

While not a traditional component of a scoping review, a critical appraisal of all individual sources of evidence was completed using the Mixed Methods Appraisal Tool 2018 (MMAT).[62] The MMAT was used to identify trends in study quality, and provide the reader insight into the quality of evidence being presented. The MMAT is a brief, five-question scale, with individual questions for five different methodological categories, allowing the tool to assess quality across study designs. Previous versions (MMAT 2011)[63] have been validated for reliability and efficiency, and the present version (2018) was revised based on feedback from MMAT users, and an e-Delphi study with international experts.[62] While not designed for formal scoring of the individual records, the MMAT provided insight into the overall methodological quality of included sources, especially within study design categories. Quality rating was reported as 0 (poor quality) to 5 (excellent

quality) based on how many of the five required elements for a study design category were included in the individual study. As recommended by the developers of the MMAT, members of the study team (CH, LT, JW) clarified the criteria for scoring through discussion and consensus prior to use. The customized MMAT criteria (Appendix E) were then piloted across multiple study design categories by two independent reviewers (CH, LT) prior to critical appraisal of all included sources by two independent reviewers (CH, LT). Any MMAT scoring discrepancies were resolved through consensus (first between the two reviewers and if required with the senior author).

4.6 Data synthesis

Following data charting, descriptive and numerical summaries of study details, sample characteristics, intervention details, and adherence concepts were consolidated. As no previous reviews on this topic in youth exist to serve as a template, a review studying adherence to exercise therapy in adults[32] was used as a template to present intervention and adherence concepts. Using a pre-existing template provided consistency in reporting and allows for future comparison of adherence concepts between youth and adults. Adherence modifiers (barriers, facilitators, and strategies to improve adherence) were grouped into prevalent topics (CH). Braun and Clarke's 6 stage guide to thematic analysis[64] was then applied to the topics to inductively identify semantic themes from these topics. Themes were then categorized into behaviour change elements using the COM-B framework (CH).[54] Theme identification and COM-B categorization was developed and trialled by the lead author (CH) and senior author (JW) until consensus was reached.

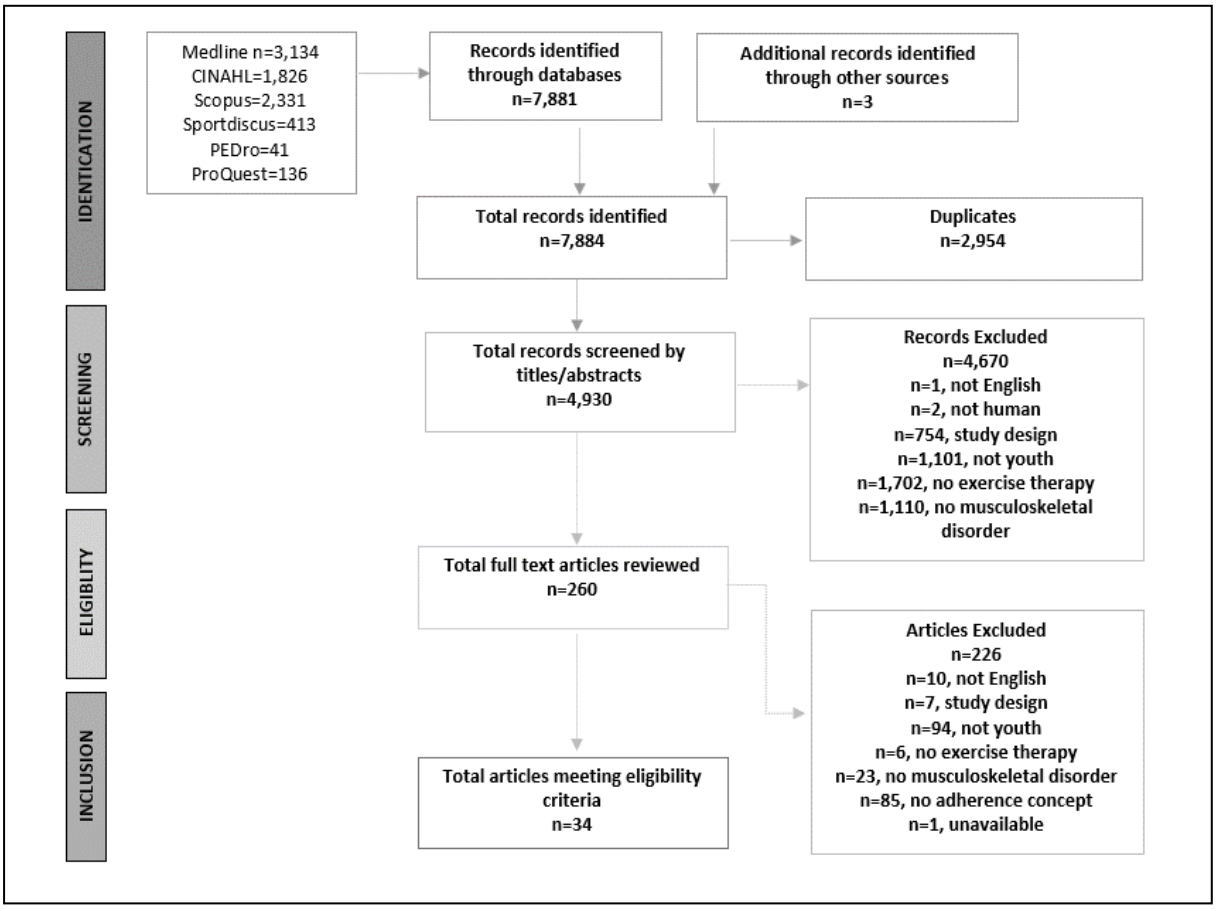
Due to their previous use in behaviour change studies, both the COM-B framework and the Theoretical Domains Framework (TDF) were considered as a guide for theme identification.[54,55] However, despite similarities between TDF domains and our identified themes, inductive thematic identification was selected as a preferred model for theme identification due to the overlap between TDF domains and lack of consistency of theme coding during pilot testing. The COM-B framework was retained to categorize identified themes as it provided consistent language and a theoretical lens through which to examine specific themes affecting behaviour. This model also provided direction and guidance to future intervention strategies, clinical implications, and helped identify potential knowledge gaps via highlighting under-researched areas.

5 Results

5.1 Study selection and characteristics

Our database search was performed in July of 2018 and yielded 7,881 potentially relevant records. Three additional records were identified through hand searching of included information sources. After removal of duplicates (n=2,954), 4,930 records were screened, 260 were reviewed in full, and 34 were included. An overview of the study selection process is presented in Figure 2.

FIGURE 2. Flowchart diagram of search results and study selection.



The characteristics of the included studies, grouped by MMAT methodological category (see Appendix E), are summarized in Appendix D. Forty-four percent (n=15) of the studies were randomized intervention studies (i.e., randomized controlled trials), 29% (n=10) were non-randomized quantitative studies (e.g., pre-experimental, cohort, cross-sectional), 9% (n=3) were quantitative descriptive studies (e.g., case series, case report), and 18% (n=6) were qualitative studies. The majority of studies were from Canada (26%, n=9), followed by the United States

(15%, n=5), Denmark (12%, n=4), United Kingdom (12%, n=4), South Africa (9%, n=3), the Netherlands (6%, n=2), Australia (6%, n=2), and 5 other countries (3%, n=1 each). With respect to study quality, seven studies (21%) were rated a 1 (poor quality), four (12%) were rated a 2, nine (26%) were rated a 3, ten (29%) were rated a 4, and four studies (12%) were rated a 5 (excellent quality). General trends across study categories found an overall poor rating with regards to internal validity, specifically as it related to administration of the assigned intervention (quantitative studies) and poor substantiation of results from the data (qualitative studies).

5.2 Sample Characteristics

Included studies represented data from 1,563 participants (65% female, n=1,010) with a median age of 13.2 years (ranging between 2 and 19 years). Musculoskeletal conditions represented include Adolescent Idiopathic Scoliosis (29%), Juvenile Idiopathic Arthritis (29%), Patellofemoral Pain Syndrome (15%), Juvenile Fibromyalgia (6%), and seven others (21%; see Appendix D). Details of exercise therapy interventions for all studies are summarized in Table 1. Across studies, prescribed exercise therapy length ranged between 4 to 160 weeks, with individual sessions ranging between 15 to 120 minutes, at a frequency spanning 2 to 21 sessions/week. Most exercise therapy programs were multimodal (e.g., strength and aerobic components), featured partial supervision (e.g., a mix of supervised and unsupervised exercise), and were completed in combined settings (e.g., home and clinic components). Exercise therapy details were fully reported in only 47% of studies.

TABLE 1. Reported characteristics of exercise therapy interventions.

Type of exercise	**n (% of studies)**
multi-modal	16 (47%)
strength	5 (15%)
aerobic	4 (12%)
motor control	3 (9%)
not reported	5 (15%)
Supervision	**n (% of studies)**
partial	17 (50%)
none	7 (21%)
full	6 (18%)
not reported	4 (12%)
Setting	**n (% of studies)**
combined	18 (53%)
home	10 (29%)
clinic/hospital	2 (6%)
not reported	4 (12%)
	Range
Prescribed session length	15-120 minutes
Prescribed session frequency	2-21 sessions/week
Prescribed program length	4-160 weeks

Reported adherence variables are summarized in Table 2. Sixty-eight percent (n=23) of studies did not report a value for minimal acceptable adherence (adherence goal). Of those that did, the minimal acceptable adherence ranged from 31 to 100% of prescribed exercises. Reported adherence across the 59% of studies that reported it (n=20) ranged between 15 to 99% POP. The most commonly used adherence outcome and measurement tool were “session completion” (32% of studies), and “self report exercise log” (53% of studies), respectively.

TABLE 2. Reported adherence rates, outcomes, and instruments across included studies.

Adherence rates	**Range**
Adherence Goal (% of prescribed) Not reported: n=23 (68%)	31-100%
Adherence Rate (% of prescribed) Not reported: n=14 (41%)	15-99%
Adherence outcome	**n (% of studies)***
Session completion	11 (32%)
Exercise frequency	9 (26%)
Exercise intensity	3 (9%)
Session attendance	2 (6%)
Exercise time	1 (3%)
Exercise replication	1 (3%)
Behaviour component	1 (3%)
Not reported	7 (21%)
Adherence instrument	**n (% of studies)***
Self report exercise log	18 (53%)
Class register	11 (32%)
Objective measure	6 (18%)
Interview	4 (12%)
Existing measurement scale	2 (6%)
Health care provider observation	1 (3%)
Not reported	7 (21%)

**Percentages may equal more than 100% as individual studies may report multiple adherence outcomes/measures*

5.3 Thematic Synthesis

A total of 190 data items related to exercise therapy adherence were charted across studies. From those, 55 unique topics were distinguished, and 11 consistent themes identified. In total, 12% of data items were related to Capability, 48% to Opportunity, and 40% to Motivation resulting in three themes related to Capability, four to Opportunity, and four to Motivation. There was substantial variability in topics identified in each study, with multiple studies identifying topics unique to that sample. The frequency of themes and topics within those categories are presented in Table 3 and discussed in detail below. Please refer to Appendix D for specific sample characteristics including age, diagnoses, and adherence instrument for all included studies.

TABLE 3. Frequency of identified themes, organized by COM-B category, with study quality ratings and topics.

COM – B Category *Theme*	**Barrier*** *Theme Frequency, Study Quality (range, 0-5), Topic*	**Facilitator*** *Theme Frequency, Study Quality (range, 0-5), Topic*	**Strategy*** *Theme Frequency, Study Quality (range, 0-5), Topic*
Capability			
Beliefs	2 (1-5) Understanding the purpose of exercise (lack of)[65,66]	1 (2) Perceived helpfulness of treatment[67]	**10** (1-4) Education on the purpose of exercise[19,68–72] Education on the consequences of non-compliance[34,50,73] Education about condition[19]
Physical Characteristics	3 (2) Fatigue[74] Other symptoms[74]	1 (2) Younger age[67]	0
Psychological Characteristics	3 (3-5) Personality traits[34] Confidence (lack of)[75] Laziness[65]	2 (3) Personality traits[34] Self confidence[76]	0
Opportunity			
Physical Environment	**12** (2-4) Transportation issues[70,75–79] Privacy (lack of)[78,80,81] Location (pool)[78] Location (home)[74]	6 (2-4) Location (home)[37,74] Location (community)[75] Location (pool)[78] Low cost[37] Equipment provided[76]	4 (2-5) Providing equipment[74,82] Minimizing cost[76] Selecting school as location[20]
Social Environment	0	**18** (1-5) Family involvement[66,75,80] Group environment[65,76] Strong therapeutic alliance[69,76,83] Comparison to others[75,76] Peer support (in person)[76] Social support[34,84] Peer support (social media)[81]	9 (1-5) Family involvement[19,20,37,69,82,85] Using an online chat forum[81] Involving friends[72] Fostering therapeutic alliance[77]
Time	**13** (2-5) Time constraints[65,74,77,79,80,83] School responsibilities[75,76,81,86,87] Non-flexible exercise schedule[81]	9 (3-5) Exercise schedule[65,75,76,80,81,83] Low exercise quantity[83,88] Time (available)[37]	2 (1-4) Reducing exercise quantity[35,70]
Program Details	3 (2-4) High exercise complexity[70] Exercise handouts (written only)[70] Exercise progression (lack of)[89]	3 (3-4) Exercise handouts (video)[81] Exercise handouts (written)[76] Low exercise intensity[79]	**13** (1-4) Regular exercise progression[19,70,72,83] Providing exercise instruction[70,72,75,83] Providing exercise handouts (video)[77,79,88] Providing exercise handouts (written)[34,74,90]
Motivation			
Engagement	1 (4) Accountability (lack of)[80]	8 (2-4) Patient choice[37,69,81,84] Patient accountability[67] Diary keeping[91] Exercise contract[81] Individualization[76]	5 (2-4) Individualizing exercise[70,74,76] Using an exercise contract[81] Allowing patient choice in exercise[20]
Experience	**12** (1-5) Pain during exercise[50,65,67,72] Boredom during exercise[65,78,80,89] Symptom relief (lack of)[78,91] Enjoyment of exercise (lack of)[66,77]	**14** (2-5) Enjoyment of exercise[65,68,78,80,83,85,89] Symptom relief[68,80] Competition[75,80,89] Pain during exercise (lack of)[78]	2 (3-4) Fostering enjoyment of exercise[75,88]
Reinforcement	2 (1-4) Feedback on exercises (lack of)[73] Supervision (lack of)[80]	8 (2-5) Clinician checking in[75,81,88] Rewards[65,79,80] Feedback on exercise[89,92]	**17** (1-5) Clinician checking in[37,75,77,79,85,93] Review of diaries[19,34,70,72,76,81] Providing rewards[72,77,79] Providing reminders[20,37]
Goals	4 (1-5) Goal setting (not patient led)[91] Prioritization (lack of)[65,86]	2 (3-4) Goal setting[75,81]	1 (4) Goal setting[19]

**Cell values represent number of times themes were identified (most frequent in bold), study quality ratings (range 0-5), and unique topics identified within themes.*

5.4 Barriers to Exercise Therapy Adherence

A variety of barriers to exercise therapy adherence were identified. These barriers have been organized into those related to someone's capability to adhere, opportunity to adhere, and motivation to adhere, based on the COM-B framework. Specific examples are provided for illustrative purposes.

5.4.1 Capability

Psychological characteristics such as emotional instability,[34] low ego strength,[34] and low confidence in one's abilities[75] (e.g., reduced task self-efficacy) were associated with non-adherence. Self reported "laziness" was reported as a barrier in one study,[65] as was fatigue,[74] "other symptoms"[74] (e.g. from other medical conditions), and a lack of understanding the purpose of the exercise.[74]

5.4.2 Opportunity

Fifty-one percent of identified barriers to exercise therapy were related to a person's opportunity to exercise. Time constraints such as school responsibilities[75,76,81,86,87] or other activities[65,74,77,79,80,83] were highlighted by 11 studies. Physical environment, including transportation issues[70,75–78,83] (especially for clinic/hospital-based interventions), a lack of privacy (during exercise or changing into a swimsuit),[78,80,81] and location (both pool[78] and home[74] settings) was a barrier in 26% of studies. Barriers associated with exercise program details included a lack of exercise progression,[89] exercises that were too complex,[81] and only providing written handouts when digital copies were available.[70]

5.4.3 Motivation

Personal experience while performing the exercise program was the most commonly identified barrier relating to motivation, with pain,[65,67,72,92] boredom,[65,78,80,89] and an overall lack of enjoyment during exercise[66,77] being identified in 26% of studies. In one study, goal setting was related with poor adherence if the youth was not the one setting the goals, or if goals were not regularly met.[91] Other barriers from this category included a lack of accountability[80] and not making exercise a priority.[65,86]

5.5 Facilitators of Exercise Therapy Adherence

A variety of facilitators of exercise therapy adherence were identified. These facilitators have been organized into those related to someone's capability to adhere, opportunity to adhere, and motivation to adhere, based on the COM-B framework. Specific examples are provided for illustrative purposes.

5.5.1 Capability

Youth with personality traits such as high self-confidence, maturity, responsibility, and emotional stability were identified as more likely to be adherent with exercise.[34] A belief that the exercises would be helpful was related with improved adherence,[67] as was a younger age, with those aged 11-13 years more likely than older adolescents to complete their exercises.[67]

5.5.2 Opportunity

Social environment was the most commonly identified facilitator, reported in 29% of studies. Group exercise programs[65,76] and involving friends or family[66,75,80] with the exercise program were repeatedly identified as being linked with higher adherence. Peer support, whether in person[76] or online,[81] was helpful, as was a strong therapeutic alliance between the participant and the clinician.[69,76,83] Home was the preferred location for exercise, especially in younger adolescents (age 12-16 years).[37] Facilitators regarding "time" were identified in 26% of studies, and included scheduling exercises during non-school hours,[65] scheduling options,[37,75,76,80,81,83] and being prescribed a fewer quantity of exercises (therefore shorter time to complete).[83,88] Program details such as reduced intensity of the program,[79] having written or video instructions provided,[76,81] and reduced financial burdens of exercise (e.g., providing equipment, reducing registration fees) were also related to improved adherence.[37,76]

5.5.3 Motivation

Exercises that were relieving of pain and/or other symptoms,[68,78,80] were fun,[65,68,78,80,83,85,89] or were modified to include other enjoyable activities,[65] were more likely to be completed. Patient engagement was a facilitator in seven studies, and included topics such as increased patient accountability,[67] patients choosing which exercises they preferred,[37,69,81,84] or diary keeping.[91] For example, one study reported young adolescents (age 10-16 years) enjoyed the "grown up aspect" of signing an exercise contract, and it motivated them to adhere.[81] Goal setting was found to be helpful,[75,81] as was clinicians checking in on exercise progress[75,81,88] (of which SMS was reported

as being the most effective[81]), and providing feedback on exercise technique.[89,92] Positive reinforcement was linked with improved adherence, and included age-appropriate rewards for adherence such as stickers and candy for younger children[65,79] or computer time for adolescents.[80]

5.6 Conflicting Evidence

There were several topics that were identified as both barriers and facilitators to adherence. Location for exercise (physical environment) had the most inconsistency, with exercising at home,[37,74] pool,[78] and in the community[75] all described as preferred locations for exercise, while home[74] and pool[78] locations were also identified as barriers. Goal setting was linked with improved adherence in two studies,[75,81] but also identified as a barrier in two individual interviews[91] (i.e., when goal setting was completed by the health professional rather than the patient, and if goals were not regularly achieved). While providing exercise handouts was generally identified as a facilitator of adherence, both written material[76] and video material[81] were found to be preferable over the other, and providing written material only when digital material was available was identified as a barrier.[70]

5.7 Strategies/Interventions Targeting Improved Adherence

A variety of strategies to improve exercise therapy adherence were identified in the included studies. These strategies have been organized into those targeting someone's capability to adhere, opportunity to adhere, and motivation to adhere, based on the COM-B framework. Specific examples are provided for illustrative purposes.

5.7.1 Capability

Interventions targeting a youth's beliefs were used in 26% of studies. These included education about their condition,[19] the purpose of exercise,[19,68–72] and the consequences of not completing their exercise therapy.[34,73,92] There were no identified interventions specifically targeting an individual's physical or psychological characteristics.

5.7.2 Opportunity

Four studies targeted the youth's physical exercise environment such as providing equipment[74,94] or reducing financial costs associated with exercising,[76] and by selecting favourable exercise locations.[20] Adaptations to the exercise program such as regular exercise progressions or

variations,[19,70,72,83] teaching of exercise technique,[70,72,75] and the provision of written[34,74,90] or video[77,79,88] instructions for reference at home were common. Reducing the frequency of exercise sessions to be completed (decreased time) was implemented in two studies[35,70] to help find a "compromise between maintaining adequate supervision and minimalizing disruption to the patients' and families' lives".[35] Family involvement[19,20,37,69,85,94] (parents and siblings) was the most common “social” strategy, however, involving friends (e.g., allowing a friend to join exercises[72]), online support groups (e.g., chatrooms[81]), and strengthening the therapeutic alliance (e.g., increased one-on-one attention through reduced patient/clinician ratios in class[77]) were also utilized.

5.7.3 Motivation

Reinforcement, whether positive or negative, was the most commonly targeted motivational theme, and was identified in 44% of studies. Examples of reinforcement included “checking in” by clinicians to ensure exercises were being completed,[37,75,77,79,85,93] monitoring/reviewing of exercise diaries by parents or clinicians,[19,34,70,72,76,81] and utilizing rewards[72,77,79] (e.g., stickers and candy) for adherent youth. Stimulating engagement of the participants in their program through allowing patient input,[20] individualization of exercises,[70,74,76] and employing exercise contracts[81] was utilized as a strategy in several studies. Goal setting was employed as an adherence strategy on only one occasion,[19] and only two studies specifically targeted the participant’s experience during exercise.[75,88] For example, in one study, “Instructors motivated the children to participate by choosing music they liked, giving imaginative names to the exercises, and creating class competitions.”[75]

5.8 Comparison across condition groups

The studies included in our review represent 11 different musculoskeletal conditions. These conditions can be categorized into rheumatological conditions (e.g., Juvenile Idiopathic Arthritis, Fibromyalgia), traumatic/atraumatic musculoskeletal injuries (e.g., low back pain, Patellofemoral Pain Syndrome), and idiopathic scoliosis. When comparing findings across these groups, there are no obvious differences or trends: the key themes of exercise experience, time, and both physical and social environments were identified across groups.

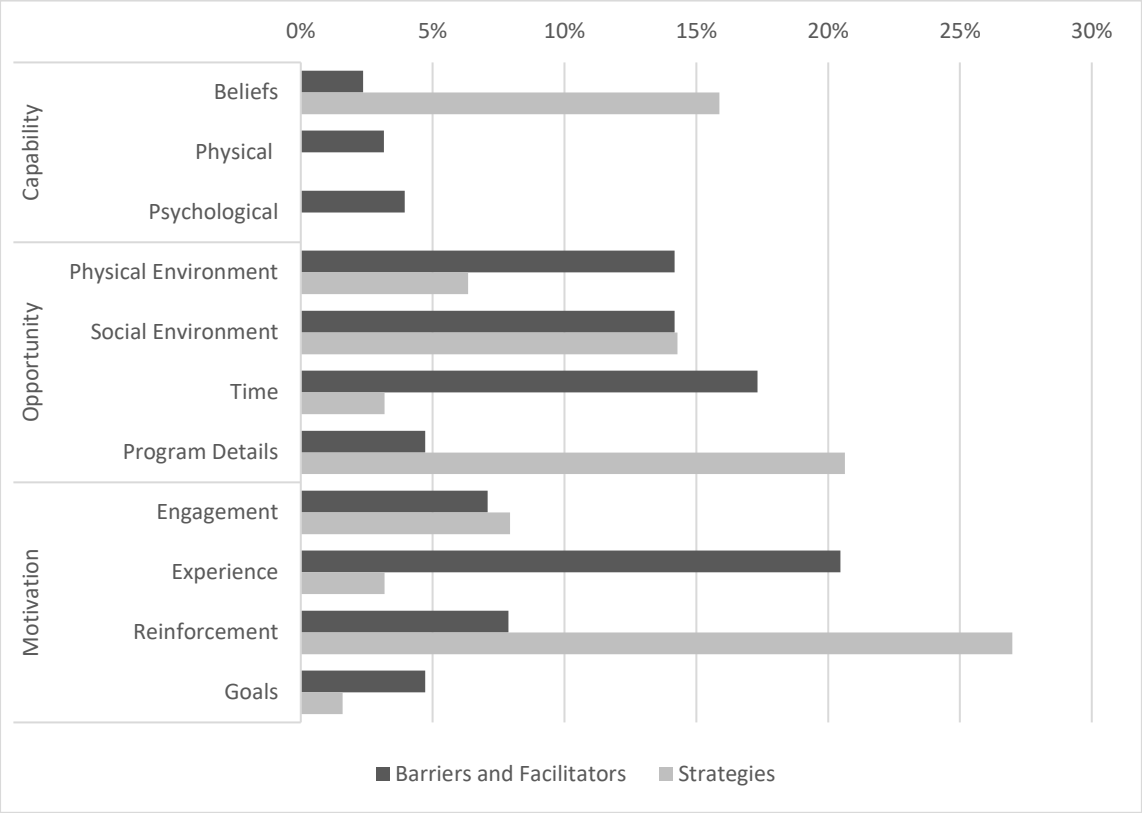
5.9 Comparison across age groups

While many of the included studies featured both children and adolescents (or did not report the age range), several studies reported findings for one group in isolation. While many of the key themes (e.g., time, environment) were represented in both age groups, there are some noteworthy differences between groups. For example, in the four studies that included only children (age ≤10 years),[69,71,79,89] two topics were heavily represented: the importance of a strong therapeutic alliance between patient and clinician,[69,79] and the importance of the ensuring exercises were enjoyable.[79,89] In contrast, the studies including only adolescents (age >10 years) repeatedly identified "school responsibilities"[75,86,94] as a barrier, and "group exercise"[76,84] as a facilitator of adherence.

5.10 Comparison between barriers/facilitators and interventions/strategies.

A summary of the themes identified as barriers/facilitators to adherence (proportion of total) vs themes targeted with interventions/strategies (proportion of total) is provided in Figure 3. The closest matched themes (i.e., where proportions of barriers/facilitators identified are similar in proportion to interventions used) included the social environment, physical and psychological characteristics, patient engagement, and goal setting. Experience during exercise, time, and physical environment were much more commonly identified as a barrier/facilitator than as a strategy. In three cases (reinforcement, beliefs, and program details), a theme was identified at a higher proportion as an intervention than as a barrier/facilitator indicating that these themes were targeted more frequently than they were identified as being impactful on adherence.

FIGURE 3. Reported barriers and facilitators (proportion of total) compared to reported adherence boosting strategies (proportion of total), organized by theme and COM-B *c*ategory.



6 Discussion

6.1 Key findings

To our knowledge, this scoping review is the first to consolidate and consider the relationship between known barriers, facilitators and boosting strategies for adherence to exercise therapy treatments for youth with musculoskeletal conditions. Few included studies reported adherence concepts, and in those that did, there was a heterogeneity of adherence outcomes and instruments employed. The most commonly reported barriers to exercise therapy programs were time constraints, physical environment, and negative exercise experience, while social environment and positive exercise experience were the most commonly identified facilitators. Reinforcement, program modification, and education were the most commonly used adherence boosting strategies, despite these themes not being commonly identified as barriers or facilitators to adherence. Several themes were largely ignored by adherence boosting strategies such as the exercise experience, time, goals, and both physical and psychological characteristics.

6.2 Comparison to other exercise therapy adherence evidence.

6.2.1 Barriers and Facilitators

Similarities exist between our findings and our current understanding of exercise therapy adherence in adults with musculoskeletal conditions. For example, experience during exercise (i.e. boredom, pain), number of exercises, time required to complete exercises, and a lack of social support have been identified as barriers to adherence in adults with musculoskeletal conditions.[44,45,95] This is understandable given that concepts of a desire for pleasure, time constraints, and importance of social interaction are not age dependent. In contrast, barriers related to transportation and school responsibilities appear unique to youth populations. Several barriers to exercise therapy in adults were not identified in our review. These included baseline physical activity,[44,45] previous non-adherence,[44,45] and psychological characteristics such as depression,[45] stress levels,[44,45] and anxiety.[45] While it is possible that these psychological characteristics are simply less impactful to youth with musculoskeletal conditions, this has not been formally studied, and evaluating psychological aspects of musculoskeletal rehabilitation in youth is an evolving field in need of further development.[96]

One important way in which our findings differ from existing evidence from adult populations was in regard to self-efficacy. While commonly associated with adherence to exercise therapy in adults,[44,45,97–100] self efficacy (defined as the “belief about one’s capabilities to produce designated

levels of performance"[101]) had minimal representation in the studies included in our review, despite being described as one of the strongest predictors of treatment adherence for childhood chronic illnesses (i.e. diabetes, asthma).[102,103] This suggests that psychological characteristics, including factors affecting intrinsic motivation (such as self-efficacy) have been possibly under studied in youth with musculoskeletal conditions.

Similarities did exist when comparing exercise therapy adherence in youth with musculoskeletal conditions to youth with other conditions. For example, rewards, family involvement, reduced time constraints, and an enjoyable exercise experience were all associated with improved exercise therapy adherence in a qualitative study of eleven adolescents with cystic fibrosis (age 10-16 years).[104] Similarly, experience during exercise, time, equipment, transportation, family/peer support, privacy, and goal setting were all identified as themes associated with adherence for youth (age ≤19 years) prescribed exercise for obesity.[105] These similarities may suggest many of the factors that influence adherence to exercise therapy in youth are related to age and social context, rather than medical condition. If true, adherence strategies found effective for other youth populations may be applicable for youth with musculoskeletal conditions and could be worth exploring.

With that said, some findings did not align across youth medical conditions. For example, while we found that social interaction consistently facilitated exercise therapy adherence in youth, a scoping review of youth with obesity found that the social environment could also be a barrier to exercise adherence.[105] Specifically, adherence was negatively impacted if friends were not active (e.g., role modeling), or if exercise evoked negative emotions related to previous activity-related teasing or bullying by peers.[105] One possible explanation for this divergence is that physical appearance and body image (a common source of social stigma) could play a larger role in youth with obesity than with musculoskeletal conditions.

6.2.2 Adherence Boosting strategies

The most commonly employed adherence boosting strategies identified in our review are consistent with those (i.e. behaviour change techniques) found in other populations. For example, reinforcement (e.g., feedback and monitoring) has been shown to be effective in improving adherence to exercise in the elderly with a variety of medical conditions.[106] For adults with persistent musculoskeletal pain, program modification (e.g., exercise practice/rehearsal), education (e.g., instruction/demonstration of behaviour), and social support all show moderate

evidence for improving adherence to exercise.[107] Patient education is also recommended to improve adherence to exercise therapy in adults with osteoarthritis,[108] despite evidence that knowledge alone does not impact behaviour change.[109]

Despite evidence of their importance in other populations, several themes were rarely targeted by adherence boosting strategies in the studies included in this review. Although goal setting is an effective behaviour change technique for improving exercise adherence in adults with musculoskeletal conditions (e.g., low back pain,[110] osteoarthritis,[111] chronic musculoskeletal pain[107]) there is a paucity of evidence of its value as an adherence boosting strategy in youth with musculoskeletal conditions.[19] Given the prevalence and effectiveness of goal setting in other populations, the lack of goal setting found in our review was surprising, especially when considering goal setting was identified as an adherence facilitator in several studies included in our review[75,81] and in studies of youth with other conditions.[104,105]

Interventions targeting improved self efficacy as an adherence boosting strategy is another area that has been shown to improve exercise therapy adherence, albeit in healthy youth,[112] but was not targeted by any study identified by our review. Given self-efficacy's previously reported association with adherence, specific self-efficacy boosting interventions may be an effective, unexplored area to target for youth with musculoskeletal conditions. Additionally, while targeting the exercise experience to help encourage adherence is popular in youth with cerebral palsy (e.g., exercise enjoyment through aquatic therapy[113] and videogame play[114]), it remained relatively ignored as an adherence boosting strategy in our review (despite it being the most commonly identified adherence predictor). Targeting the exercise experience to improve adherence would appear to be an easily adopted and fruitful area to investigate for youth with musculoskeletal conditions.

6.3 Specific population considerations

Our review included samples of diverse ages and musculoskeletal conditions. When comparing between age and condition groups, there were several population specific trends of note. For instance, when the studies were divided into condition groups (i.e. Rheumatic conditions vs idiopathic scoliosis vs traumatic/atraumatic injury), there were no obvious differences in adherence modifiers between groups. This finding would suggest that the barriers and facilitators of exercise therapy adherence in youth with musculoskeletal conditions are not dependent on condition type. If true, both existing and future adherence boosting strategies are possibly

transferrable across musculoskeletal conditions, and successful adherence interventions for one group may be applicable in others. However, it is important to note that not all musculoskeletal conditions are represented in this review, so further research is required to ensure generalizability.

Given the wide range of ages included in this review (2 – 19 years), it is important to consider factors may differ between children and adolescents. Our review did find several interesting trends when comparing findings by age group: children (≤10 years) vs adolescents (>10 years). For example, school work was a larger barrier in older adolescents than children,[75,86,94] an understandable finding given the increased homework and independent learning demands of secondary school compared to primary. Another example includes the role of the social environment in adherence. While both are forms of social support, the importance of a therapeutic alliance between patient and practitioner was highlighted in children[69,79] while group support was featured in adolescence.[76,84] This subtle difference is interesting, and possibly related to who different age groups turn to for support first (e.g., adult role models vs peers), and is worthy of future investigation. These findings are possibly meaningful for clinicians and researchers prescribing exercise therapy to one group or the other, but also emphasize the importance of individualization of adherence boosting strategies to the patient (or group) in question.

Another important consideration regarding age and adherence is caregiver influence. It is reasonable to assume that in many cases, there may be more caregiver involvement in treatment for children, than for adolescents, as independence often grows as we age (for some, more than others). Feldman et al[67] identified that younger participants were more likely than adolescents to adhere to their program. However, as there is no mention or description of caregiver involvement reported (as in the majority of studies), it is impossible to tell if these younger patients were more "adherent" or rather more "compliant" because an adult caregiver was making choices on their behalf. This observation speaks to the possible role of caregiver involvement, but also to the lack of consistent adherence reporting/measurement observed in our review and the uncertainty that creates.

Despite the possibility of caregiver involvement, many of the identified barriers, facilitators, and strategies identified by this review are supportive of the concept of patient-therapist collaboration (not *parent*-therapist) to facilitate behaviour change (i.e., the "nature of adherence"). For example, individualization,[70,74,76] patient preference,[37,69,81,84] increased accountability,[67,80] and patient-led goal setting[19,75,81,91] all reinforce the importance of *patient* participation in the pursuit of exercise

uptake. In our review, these concepts were identified in samples as young as eight years old,[69,74,84] suggesting that patient collaboration and individualization are important even in young age groups, and could be impactful when trying to improve adherence for youth with musculoskeletal conditions. These findings also support the argument for choosing “adherence” over “compliance” when discussing exercise completion to reinforce the importance of collaboration when facilitating behaviour change in this population.

6.4 COM-B implications

The use of the COM-B framework in this review provides interesting insight into the areas that adherence research has focused on. For instance, most barriers and facilitators identified in our review related to the COM-B categories of someone’s Opportunity to adhere and someone’s Motivation to adhere. Developing adherence boosting strategies focusing on these areas would seem prudent and should target related themes (such as the exercise experience, minimizing time constraints, physical/social environments, and possibly goal setting). However, given the importance of psychological characteristics to adherence in other populations, the minimal themes relating to someone’s Capability to adhere in this review could indicate an under-researched area. Future researchers should consider capability when investigating adherence, and strategies targeting capability (including self-efficacy and knowledge) should be explored. Given our findings, as all three categories play an important role in behaviour change, consideration should be given to all three during intervention planning.

6.5 Clinical implications

The findings of this scoping review highlight several areas for clinicians hoping to improve adherence when prescribing exercise therapy to youth with musculoskeletal conditions to explore. Overall, making exercise social (e.g., including friends, family, or peers), convenient (both in location and required time), and fun (e.g., using games and activities) appear important for maximizing adherence to exercise therapy for this population. However, given the variety of barriers and facilitators reported across studies, it is essential that clinicians take the time to identify the individual barriers and facilitators for each patient. Specific boosting strategies can then be selected for the individual, targeting those themes (i.e., matching barriers/facilitators and boosting strategies). Table 4 provides practical examples of adherence boosting strategies targeting the key themes identified in this review. In addition, open discussion with the patient and family at the time of exercise therapy prescription can provide clinicians the opportunity to problem

solve potential barriers and embodies the collaborative nature of adherence. Discussing the importance of adherence, defining what is an acceptable adherence level for that patient, and devising a measurement/monitoring strategy to evaluate exercise completion, are also important steps often overlooked early in the treatment process.

TABLE 4. Practical examples of adherence boosting strategies for commonly identified themes.

Theme	Topic	Practical example of adherence boosting strategy
Time	Exercise quantity	Consider limiting number of exercises to minimize time required.[35,70,79,88]
	Exercise schedule	For supervised sessions, consider having multiple options for class times including evenings and weekends[75,79]
Exercise Experience	Fun (children)	Consider using games and crafts during exercise[65]
		Incorporate preferred and recognizable activities into exercises (e.g., favorite sport)[88]
	Fun (adolescents)	Suggest adolescents select their own motivating music during exercise[75]
		Consider making exercises into games or competition[75,80,89]
		Incorporate technology/social media into the exercise routine[81]
	Pain	Check in often to ensure exercises are comfortable[78]
	Boring	Progress or change exercises regularly to prevent monotony[89]
		When possible, consider distraction techniques during exercise including books, games, and technology[89]
Social Environment	Family	Invite parents and/or siblings to participate in exercises (e.g., family challenge)[80]
	Friends	Invite friends to a participate in exercises (e.g., bring a friend to physio day)[72]
	Therapeutic alliance	Establish good therapeutic alliance early through interaction and colloaboration.[69]
		For group exercise classes, consider reducing the therapist-participant ratio for more one-on-one attention[77,79]
	Group	Consider group exercise classes for similar conditions/ages[65,76,84]
		Involve school teachers to possibly incorporate exercise into classroom routines[65]
	Online	Seek out condition specific online forums or chatrooms for youth[81]
Physical Environment	Location	Design programs that are not dependent on location for completion[20]
		Consider unconventional locations for exercise routine (e.g., school, park, pool)[20,65,78]
		For supervised exercise, offer multiple locations for sessions, and consider factors such as proximity to home and parking[20,70,79]
	Equipment	When possible, provide equipment or design programs with minimal equipment requirements[74,82]

6.6 Strengths and limitations

Despite the use of established frameworks, reporting guidelines, and rigorous methodology, there are limitations to our review. It is possible that not all barriers and facilitators to exercise therapy adherence are identified given the methodological limitations of the existing evidence base. It is also possible that sources excluded due to study design (e.g., editorials, blogs, etc.) may have contributed additional information related to these topics. Despite using piloted data charting tools, theme misclassification is possible due to overlap between categories. While study quality was evaluated and reported, studies were not excluded based on quality rating, possibly impacting the validity of reported findings. In spite of including a wide variety of conditions, ages, and interventions, generalizability of our findings may be limited as not all musculoskeletal conditions were reflected in our review. A key limitation was that formal stakeholder (i.e. clinician, patients, caregivers) consultation did not take place as part of this review. However, the study team did comprise multiple clinicians who were able to add that perspective to the interpretation. Lastly, emphasis has been placed on the frequency of reported themes in this review, however it is important to note that frequency does not necessarily reflect the importance of certain themes on adherence, purely how often they have been investigated or identified.

6.7 Future directions

Several future research steps have been identified in order to improve adherence to exercise therapy in youth with musculoskeletal conditions. A preliminary step for this process, described as mapping out or identifying predictors of adherence/non-adherence (e.g., barriers and facilitators),[38] has been addressed through this review. However, given the variety of methods to measure adherence and overall lack of reporting of adherence concepts identified by our review, it is clear that a standardized approach for both measuring and reporting adherence concepts is needed. At minimum, consistent use of terminology related to adherence and compliance should be respected, and the reporting of the percentage of prescribed exercises completed (regardless of measurement tool) would allow for comparison and pooling of results across studies. Considering the consistency of adherence concepts identified across condition and age groups, future research should focus on not replicating this work, rather developing screening tools to identify known barriers and facilitators at an individual level, and assessing the effect of interventions that target individualized adherence boosting strategies on adherence and clinical outcomes.

7 Conclusion

Despite adherence concepts being poorly defined and reported across studies, a variety of barriers, facilitators, and strategies impacting exercise therapy adherence were identified for youth with musculoskeletal conditions. Time constraints, physical environment, and negative exercise experiences were commonly identified barriers, while social support and positive exercise experiences were frequently identified facilitators. Reinforcement, exercise program modification, and education were commonly used adherence boosting strategies, despite being infrequently reported as barriers or facilitators. Efforts to match adherence boosting strategies to an individual's needs should be considered and should consider the key themes of exercise experience (positive/negative), time, and environment (physical and social) to maximize effectiveness. Consensus on adherence measurement and reporting is required prior to the development of adherence screening tools and individualized adherence interventions. Based on evidence from other populations, targeting psychological characteristics (such as self-efficacy) and goal setting techniques should also be considered as areas of future investigation.

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Appendices

Appendix A: PRISMA Extension for Scoping Reviews, as per Tricco *et al* (2018)[59]

Table. PRISMA-ScR Checklist

Section	Item	PRISMA-ScR Checklist Item
Title	1	Identify the report as a scoping review.
Abstract		
Structured summary	2	Provide a structured summary that includes (as applicable) background, objectives, eligibility criteria, sources of evidence, charting methods, results, and conclusions that relate to the review questions and objectives.
Introduction		
Rationale	3	Describe the rationale for the review in the context of what is already known. Explain why the review questions/objectives lend themselves to a scoping review approach.
Objectives	4	Provide an explicit statement of the questions and objectives being addressed with reference to their key elements (e.g., population or participants, concepts, and context) or other relevant key elements used to conceptualize the review questions and/or objectives.
Methods		
Protocol and registration	5	Indicate whether a review protocol exists; state if and where it can be accessed (e.g., a Web address); and if available, provide registration information, including the registration number.
Eligibility criteria	6	Specify characteristics of the sources of evidence used as eligibility criteria (e.g., years considered, language, and publication status), and provide a rationale.
Information sources*	7	Describe all information sources in the search (e.g., databases with dates of coverage and contact with authors to identify additional sources), as well as the date the most recent search was executed.
Search	8	Present the full electronic search strategy for at least 1 database, including any limits used, such that it could be repeated.
Selection of sources of evidence†	9	State the process for selecting sources of evidence (i.e., screening and eligibility) included in the scoping review.
Data charting process‡	10	Describe the methods of charting data from the included sources of evidence (e.g., calibrated forms or forms that have been tested by the team before their use, and whether data charting was done independently or in duplicate) and any processes for obtaining and confirming data from investigators.
Data items	11	List and define all variables for which data were sought and any assumptions and simplifications made.
Critical appraisal of individual sources of evidence§	12	If done, provide a rationale for conducting a critical appraisal of included sources of evidence; describe the methods used and how this information was used in any data synthesis (if appropriate).
Summary measures	13	Not applicable for scoping reviews.
Synthesis of results	14	Describe the methods of handling and summarizing the data that were charted.
Risk of bias across studies	15	Not applicable for scoping reviews.
Additional analyses	16	Not applicable for scoping reviews.
Results		
Selection of sources of evidence	17	Give numbers of sources of evidence screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally using a flow diagram.
Characteristics of sources of evidence	18	For each source of evidence, present characteristics for which data were charted and provide the citations.
Critical appraisal within sources of evidence	19	If done, present data on critical appraisal of included sources of evidence (see item 12).
Results of individual sources of evidence	20	For each included source of evidence, present the relevant data that were charted that relate to the review questions and objectives.
Synthesis of results	21	Summarize and/or present the charting results as they relate to the review questions and objectives.
Risk of bias across studies	22	Not applicable for scoping reviews.
Additional analyses	23	Not applicable for scoping reviews.
Discussion		
Summary of evidence	24	Summarize the main results (including an overview of concepts, themes, and types of evidence available), link to the review questions and objectives, and consider the relevance to key groups.
Limitations	25	Discuss the limitations of the scoping review process.
Conclusions	26	Provide a general interpretation of the results with respect to the review questions and objectives, as well as potential implications and/or next steps.
Funding	27	Describe sources of funding for the included sources of evidence, as well as sources of funding for the scoping review. Describe the role of the funders of the scoping review.

JBI = Joanna Briggs Institute; PRISMA-ScR = Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews.
* Where *sources of evidence* (see second footnote) are compiled from, such as bibliographic databases, social media platforms, and Web sites.
† A more inclusive/heterogeneous term used to account for the different types of evidence or data sources (e.g., quantitative and/or qualitative research, expert opinion, and policy documents) that may be eligible in a scoping review as opposed to only studies. This is not to be confused with *information sources* (see first footnote).
‡ The frameworks by Arksey and O'Malley (6) and Levac and colleagues (7) and the JBI guidance (4, 5) refer to the process of data extraction in a scoping review as data charting.
§ The process of systematically examining research evidence to assess its validity, results, and relevance before using it to inform a decision. This term is used for items 12 and 19 instead of "risk of bias" (which is more applicable to systematic reviews of interventions) to include and acknowledge the various sources of evidence that may be used in a scoping review (e.g., quantitative and/or qualitative research, expert opinion, and policy documents).

Appendix B: Search Strategy and Results

For all databases, Medical Subject Headings (MeSH) and keywords were selected based on consultation with a librarian scientist and content experts from amongst the study team. Truncation and alternate spelling (i.e., North American AND European) were utilized as appropriate for all keywords, while subject headings were selected from available options, varying by database.

Ovid MEDLINE

1. exp Pediatrics/ or exp Adolescent/ or exp Child/
2. adolescen* OR youth* OR teen* OR child*
3. 1 or 2
4. exp Patient Compliance/ or exp Compliance/ or exp Patient Participation/ or exp Treatment Adherence/
5. adher* OR adhear* OR compliance OR fidelity OR participa* OR barrier* OR facilitator* OR behaviour change OR behavior change OR adopt* OR uptake OR congruence OR maintenance
6. 4 or 5
7. exp Exercise Therapy/ or exp Exercise Movement Techniques/ or exp Physical Conditioning, Human/
8. exercise therap* OR therapeutic exercise* OR exercise program* OR home program*
9. 7 or 8
10. 3 and 6 and 9

CINAHL

1. (MH "Adolescence+") OR (MH "Child+") OR (MH "Pediatrics+")
2. adolescen* OR youth* OR teen* OR child*
3. 1 or 2
4. (MH "Patient Compliance+") OR (MH "Behavioral Changes+")
5. adher* OR adhear* OR compliance OR fidelity OR participa* OR barrier* OR facilitator* OR “behaviour change” OR “behavior change” OR adopt* OR uptake OR congruence OR maintenance

6. 4 or 5
7. (MH "Therapeutic Exercise+")
8. “exercise therap*” OR “exercise program*” OR “therapeutic exercise*” OR “home program*”
9. 7 or 8
10. 3 and 6 and 9

Scopus

1. adolescen* OR youth* OR teen* OR child*
2. adher* OR adhear* OR compliance OR fidelity OR participa* OR barrier* OR facilitator* OR “behaviour change” OR “behavior change” OR adopt* OR uptake OR congruence OR maintenance
3. “exercise therap*” OR “exercise program*” OR “therapeutic exercise*” OR “home program*”
4. 1 and 2 and 3

SPORTDiscus

1. (DE "CHILDREN") OR (DE "YOUTH") OR (DE "TEENAGERS") OR (DE "PEDIATRICS")
2. adolescen* OR youth* OR teen* OR child*
3. 1 or 2
4. DE “EXERCISE adherence”
5. adher* OR adhear* OR compliance OR fidelity OR participa* OR barrier* OR facilitator* OR “behaviour change” OR “behavior change” OR adopt* OR uptake OR congruence OR maintenance
6. 4 or 5
7. DE "EXERCISE therapy" OR DE "EXERCISE therapy for children"
8. “exercise therap*” OR “therapeutic exercise*” OR “exercise program*” OR “home program*”
9. 7 or 8
10. 3 and 6 and 9

PEDro

1. “children adherence exercise”
2. “adolescent adherence exercise”
3. “children adherence home program”
4. “adolescent adherence home program”

ProQuest

1. noft(adolescen* OR youth* OR teen* OR child*) AND noft(adher* OR adhear* OR compliance OR fidelity OR participa* OR barrier* OR facilitator* OR "behaviour change" OR “behavior change” OR adopt* OR uptake OR congruence OR maintenance) AND noft("exercise therap*" OR "therapeutic exercise*" OR "exercise program*" OR "home program*")

Results

Database	**Date Searched**	**# of Results**
MEDLINE	June 28, 2018	3,134
CINAHL	June 28, 2018	1,826
Scopus	June 28, 2018	2,331
SPORTDiscuss	June 28, 2018	413
PEDro	June 28, 2018	41
ProQuest	June 28, 2018	136
	Total	**7,881**

Appendix C: Exclusion Criteria with Justification

1. **English:** English language only. This restriction was in place to help keep the search feasible. Formal translation of all identified non-English records was not available for this study due to time and budget restraints
2. **Human:** Records include human subjects. Animal studies were not applicable or informative for this research question.
3. **Study Design:** Only records of experimental, quasi-experimental, observational and qualitative study design with original data were included. Reviews, editorials, commentaries, and conference proceedings were not included as they were unlikely to provide original data.
4. **Youth (aged ≤19 years) Sample:** The study sample included youth between the ages of ≤19 years. If the sample included a mix of adults and youth, pertinent data from youth samples was clearly separated from adults and extractable. This age limit was chosen to reflect all children and "school-aged" adolescents and was iteratively increased from ≤18 years during title/abstract screening as multiple informative records with samples including youth aged 19 years were identified.
5. **Adherence Construct:** All included records identified or discussed a barrier or facilitator to exercise therapy adherence, or a strategy/intervention to improve adherence to exercise therapy.
6. **Exercise Therapy Intervention:** Included records contained an exercise therapy intervention. Exercise therapy was defined as "a regimen or plan of physical activities designed and prescribed for specific therapeutic goals whose purpose is to restore normal musculoskeletal function or to reduce pain caused by diseases or injuries".[16] Records examining general exercise guidelines such as general physical activity recommendations were not included.
7. **Musculoskeletal Condition:** Intervention must have been in relation to a musculoskeletal condition or injury. Healthy participants participating in general exercise programs were not included. Malignancy of the musculoskeletal system (e.g., osteosarcoma), neuromuscular conditions (e.g., Duchenne's Muscular Dystrophy), injuries to the skin and fascia (e.g., thermal burns), and obesity were also excluded as findings from these samples would be not be generalizable due to their unique rehabilitation/exercise therapy contexts.

Appendix D: Data Charting Tool

Study Features (author, year, country, design, quality rating*), by study design category	Sample (MSK condition, sample size, sex, age)	Aim of Study	Intervention Details (type of exercise, supervision, sessions/week, minutes/session, program length)	Adherence Concepts (rate (POP), target (POP), measurement tool, outcome)	Barriers to Adherence (theme†, data item)	Facilitators to Adherence (theme†, data item)	Strategies to Improve Adherence (theme†, data item)
Qualitative							
De Monte *et al.* 2009 (Australia) Phenomenology 5/5	Juvenile Idiopathic Arthritis n=17 F=11 (85%) 11±N/R (8-16)	to (1) understand the children's perspective of living with JIA and (2) understand how this had an impact on their participation in home exercise programmes.	N/R N/R N/R N/R N/R	N/R N/A N/R N/R	1. Lack of understanding on the importance of exercise/long term benefits 11. A number of children (n = 7) commented that they had better things to do than exercises: 6. Time was another significant issue for not participating in exercises. It was reported that both being too busy with other things and having too many exercises to complete were considerations 3. One child reported that, 'Because I'm just too lazy' (Lisa, 12 years), she did not get the required exercises done. 9. Boredom with the repetitive nature of some exercises was a common theme that made them less appealing to do. 9. The children discussed the pain caused by arthritis, which had an impact on their participation in home exercise programmes	5.. The group setting was identified as a positive experience to meet other children with arthritis and a fun medium to enable the completion of exercises. 6. For some children (n = 3), their exercise programmes were incorporated into their school routines with the assistance of a teacher aide. 9. Another reported facilitator of exercises was making them fun. 10. Finally, rewards, such as a sticker and lollies, appeared to be a motivator for many children to complete their exercises. Although the children reported they did not get these all the time, it became apparent that they were given them at times when participation was limited to improve motivation.	N/A
Evans and Hardy. 2002 (UK) Qualitative Case Study 4/5	Sports Injuries (Torn ACL, Dislocated Shoulder) n=2 M=2 (100%) 19±0 (19-19)	The purpose of the present study was to derive cross-case summaries from case study profiles to enhance the interpretability and meaningfulness of the findings emerging from the goal-setting intervention study	Mixed N/R N/R N/R N/R	N/R N/R N/R N/R	11. Goal Setting (self selected vs surgeon), 11. Goal setting (de-motivating if was not successful) 9. Slow progress = frustration	8. Diary keeping increased confidence	N/R
Birt *et al.* 2014 (UK) Phenomenology 4/5	Joint hypermobility n=19 F=10 (53%) 12.6±N/R (9-17)	to enhance understanding of the factors underlying concordance with a multidisciplinary treatment programme for joint hypermobility in children	Mixed none 7/week N/R 12 weeks	N/R N/A N/R N/R	8. Child not taking responsibility for competing exercise 10. Unsupervised setting 6. Lack of time and physical resources 4. Lack of privacy 9. Monotony of exercise	5. Parent Supervision 6. Building the exercises into daily family routines. 10. Activities as consequential rewards for completion: 9. Fun exercises 9. Competition with others (family) 5. Completing exercises together with family 9. Experienced a physical improvement, which in turn created motivation to continue with the exercises	N/R
Hutzal *et al.* 2009 (Canada) Phenomenology 4/5	Juvenile Idiopathic Arthritis n=4 F=4 (100%) N/R	to identify elements of a successful community-based exercise program for children with arthritis by investigating the perspectives of fitness instructors who led the program	Aerobic Partial 3/week 45-55 minutes1 2 weeks	N/R N/R N/R N/R	3. Children who could not keep up with their healthy peers often enteredthe program with little confidence in their physical abilities 6. the children had homework burden and social responsibilities that acted as obstacles to integrating the exercise program into their lives. 4. They relied on family members to take them to class and encourage them to exercise at home. This assistance was often limited by family routines.	6. exercise schedule, integrating exercise into their lifestyle 10. Follow up in class or on phone11. formation of activity goals 4. Community centres were better than hospitals for this type of program. 5.. Parental and Peer SupportParental encouragement seemed to influence children's participation in class and at home. 9. healthy sense of competition during the classes and some 5. children were inspired by the sports and activities others with JIA performed.	7. The instructors taught children techniques to help them participate maximally in the sessions, such as decreasing the excursion in sore joints, reducing exercise intensity, changing the pace, or avoiding exercising painful joints 10. The instructors felt that close monitoring and feedback were essential tochildren's success 9. Instructors motivated the children to participate by choosing musicthey liked, giving imaginative names to the exercises, and creating class competitions.

Kashikar-Zuck *et al.* 2016 (USA) Qualitative Case Study 3/5	Juvenile Fibromyalgia n=17 F=17 (100%) 16±2.15 (12-18)	to 1) obtain information about the feasibility, safety, and tolerability of the 8-week (16 session) group-based FIT Teens intervention for adolescents with JFM and 2) gather detailed feedback from participants about their impressions of the acceptability, format, and content of the program	Motor Control full 2/week 30 minutes 8 weeks	N/R N/R Class register Session Attendance	6. Timing of session (unwillingness to miss school) 4. Transportation issues	5. (Group) group format allowed to motivate one another 3. Not just being forced to do traditional physical therapy… with this being catered towards people with fibromyalgia. I was thinking that if she can do it then I can do it too. 6. I need some sort of a plan. So this was helpful how it is always scheduled same day, same time. 5. (Therapeutic Alliance) I get the concept but I forget the positioning exactly. So then they [exercise physiologist and psychologist] would right away be like ok here. It was never like, "Why would you forget something like that?"…they were really helpful. 4. + 7. At the end of it she gave us this program [handouts] so we can keep on going…I think I'll do really well with them. We got the BOSU [balance trainer device] and … a packet of exercises [to take home]. 8. Nearly all participants reported that the pace and progression of learning exercises was a positive feature, and the majority reported that the interventionists appropriately modified the exercises as needed to meet individual levels of ability. 5. (Group) Participants were unanimously positive about the group format, mainly due to the supportive and encouraging group environment and the opportunity to meet other patients with JFM (Table 6). 5. (Peer Support)I really liked being with the other girls… I never knew they were going through the same things or… the same problems… knowing someone else feels the same way you do really helped. 5. (Peer Support) We were able to see how they were doing [the exercises] and encourage each other. The support was the biggest thing. 5. (Peer Support) Getting to know other people with fibromyalgia around my age was nice. 5. (Group) I liked learning the exercises in a group format…I liked knowing that I wasn't the only one confused or I wasn't the only one that wasn't that excited about learning this really complicated exercise. 5. (Comparison) [I] needed that re-assurance for myself…I saw that the other girls were fine with [exercises] I was like ok you are worrying over nothing. 5. (group) [I] wouldn't want to be doing it on my own… because you wanna have other people in the group cause you know that there's other people out there that are just like you and you also learn from their experience.	8. Specific tailoring of program to individual 10. Review of diaries 4. Removal of financial barriers (reimbursement of transportation and class expenses)
Williams *et al.* 2015 (UK) Qualitative Case Study 3/5	Adolescent Idiopathic Scoliosis n=6 F=6 (100%) 14.3±2.25 (10-16)	To explore factors that influenced the acceptability and perception of the trial and interventions, issues influencing exercise adherence and appropriateness of the chosen outcome measurement to participants of the exercise programme.	Motor Controlnone7/weekN/R26 weeks	56% N/R Self report exercise log Online Exercise diary (monitored) Exercise Frequency	6. Work hours of physios (supervisors)6. The children described the need to adjust their busy school routine to fit in the exercise 7. complexity of exercise program difficult for children to "grasp", however the children did not describe the exercises as difficult to grasp. 4. lack of private space to exercise	6. flexibility of schedule for supervised sessions 7. reminder materials: Children found the online diary useful as a reminder, particularly the videos. They confirmed that a paper copy could be a useful backup. 10. 'someone checking up on me' (parent or physiotherapist) was an incentive to keep going. (SMS text messaging was most helpful) 8. Signing a contract helped motivate some children to participate ("Grown up aspect" was enjoyable) 11. goal setting was possibly helpful, but many children did not remember participating 5. Peer support through existing social media sites was possibly helpful 8. Patient choice	5. Online chat forum (social support, access to clinicians for questions) 8. Exercise Contract 10. Monitored exercise diary
Quantitative Randomized							
Mendoca *et al.* 2013 (Brazil) RCT 5/5	Juvenile Idiopathic Arthritis n=50 F=32 (64%) INT: 11.0±3.9 (N/R) CNT: 11.8±3.4 (N/R)	to assess the effects of a Pilates exercise program on HRQOL in individuals with JIA.	Mixed full 2/week 50 minutes 24 weeks	CNT: 95% INT: 99% N/R Self report exercise log (attendance frequency) session attendance	N/A	9. "Pilates exercises are more motivating" than traditional exercise	10. Physiotherapists had frequent contact with patient's caregivers to encourage adherence 5.. Caregiver involvement

Schreiber *et al.* 2016 (Canada) RCT 5/5	Adolescent Idiopathic Scoliosis n=50 F=47 (94%) 13.4±1.6 (N/R)	to determine the effect ofa six-month Schroth PSSE intervention added to standard ofcare (observation or bracing) on the Cobb angle, compared to the standard ofcare alone in patients with AIS.	Mixed partial 8/week Supervised: 60 minutes Unsupervised: 30-45 minutes 26 weeks	Supervised: 85% Unsupervised: 82.5% 70% of prescribed Class register, self report exercise log (logbook) Session attendance, Exercise frequency	6. time constraint due to homework	N/R	N/R
Schreiber *et al.* 2015 (Canada) RCT 5/5	Adolescent Idiopathic Scoliosis n=50 F=47 (94%) 13.4±1.6 (N/R)	to determine the effect of a 6-month Schroth exercise intervention in conjunction with standard of care (observation and bracing) on QOL, perceived appearance and back muscle endurance, compared to the standard of care alone in patients with adolescent idiopathic scoliosis	Mixed partial 8/week Supervised: 60 minutes Unsupervised: 30-45 minutes 26 weeks	Supervised: 85% Unsupervised: 82.5% 70% of prescribed class register, self-report exercise logs (monitored) session attendance, exercise frequency	N/R	N/R	4. provided home equipment, access to facilities, and 5.. promoted parental involvement. ("When compliance dropped below 70 %, we tried to resolve the issues cooperatively with patients and parents")
Wiegerinck *et al.* 2016 (Netherlands) RCT 4/5	Calcaneal Apophysitis n=101 F=25 (25%) 10.6±1.6 (N/R)	to compare the effectiveness of 3 conservative treatment strategies for calcaneal apophysitis, with decrease of pain being the primary outcome measure.	Strength partial 7/week N/R 12 weeks	N/R N/R N/R N/R	N/R	N/R	1. Clearly explaining the expected results at onset
Tarakci *et al.* 2012 (Turkey) RCT 4/5	Juvenile Idiopathic Arthritis n= 93 F=37 (46%) INT: 10.02±3.44 (5-17) CNT: 10.82±4.00 (5-16)	to investigate the effects of an individually planned land-based home exercise (LBHE) programme on pain, functional ability, and quality of life, using a randomized, controlled, single-blind design.	Mixed Full 4/week 20-45 minutes 12 weeks	≥ 75% N/R Self report exercise log (Monitored exercise diary for completion) exercise frequency	4. transportation difficulty (geographic location too far from hospital) 7. paper handout only (no videos provided)	N/R	7. gradual increase of quantity of ex program (# of ex, repetitions, total time) 6. Reduced number of exercises7. Demonstration by Physical Therapist8. Individualization of program1. Education to patient and parent (unexpected responses with respect to exercise training and joint protection, how the recommended exercise programme may help them, and specific information about how to exercise safely and effectively and how to recognize post-exercise soreness).10. compliance diary reviewed weekly
Fanucchi *et al.* 2009 (South Africa) RCT 4/5	Low back pain n=72 F=33 (46%) INT: 12±0.7 (12-13) CNT: 12±0.7 (12-13)	To determine if an eight-week exercise program would reduce the intensity and prevalence of low back pain in 12–13 year old children, and would it decrease the childhood physical risk factors for low back pain and promote a sense of well-being.	N/R partial 4/week Supervised: 40-45 minutes Unsupervised: N/R 8 weeks	Supervised: 87% Unsupervised: 33% Supervised = Full attendance, Unsupervised = ≥3/week Class register self report exercise log session attendance exercise frequency	N/R	9. Enjoyment 9. Symptom relief ("Felt that the exercises helped to make them feel better") 9. Functional improvement ("make their backs feel stronger")	1. A physiotherapist discussed the importance of the exercises which the children would be doing and how the exercises related to their low back pain.
Singh-Grewal *et al.* 2007 (Canada) RCT 4/5	Juvenile Idiopathic Arthritis n=80 F=64 (80%) INT: 11.7±2.5 (8-16) CNT: 11.5±2.4 (8-16)	to examine the effectiveness of a high-intensity 12-week program in terms of VO2submax in children with inflammatory arthritis and to determine the effectiveness of this program in terms of self-reported physical function, VO2peak, and peak power.	Aerobic partial 3/week 45-55 minutes 12 weeks	INT=56%, CNT=78% attended >70% of training sessions and achieved heart rates of >75% of max heart rate in >50% of sessions Self report exercise log (Diary), Obj measure (Intermittent heart rate monitor) Session completed Exercise intensity	6. Lack of time	7. Less intense exercise regimen	10. instructors maintained frequent contact 10. Children were rewarded with stickers for completed sessions and were able to trade these for small gifts 7. Videotaped instructions

Monticone *et al.* 2014 (Italy) RCT 4/5	Adolescent Idiopathic Scoliosis n=110 F=80 (73%) INT: 12.5±1.1 (N/R) CNT:12.4±1.1 (N/R)	to compare an innovative outpatient programme combining active self-correction, task-oriented exercises and education with a routinely followed programme of traditional exercises to verify whether it could reduce spinal deformities and improve HRQL in adolescents with mild AIS	Mixed partial 3/week Supervised: 60 minutes Unsupervised: 30 minutes Until maturity (approx 40 months/160 weeks)	N/R N/R Self report exercise log (diary) Sessions completed	N/R	N/R	10. Diary checked weekly 5. Parent Support 1. Education about condition 1. Education on purpose of exercise 7. Graded exposure to exercise/activity 11. Shared goal setting
Epps *et al.* 2005 (UK) RCT 4/5	Juvenile Idiopathic Arthritis n=78 F=43 (55%) INT: 12±N/R (6-19) CNT: 11±N/R (4-19)	To compare the effects of combinedhydrotherapy and land-based physiotherapy(combined) with land-based physiotherapy only on cost, health-related quality of life andoutcome of disease in children with juvenile idiopathicarthritis (JIA).	Hydrotherapy Full 8/week 60 minutes 10 weeks	Percentage: N/R "Acceptable": N/R N/R N/R	Barriers to Hydrotherapy: 4. didn't like chlorine. 6.found the hydrotherapy pool inconvenient to travel to 9. didn't feel that it worked 4. equipment/facility problems 3. changing into swimsuit 9. Boring	Facilitators for Hydrotherapy: 4. easier than land based ex 9. less painful than land based ex 9. fun and enjoyable	N/R
Stephens *et al.* 2008 (Canada) RCT (Pilot) 4/5	Fibromyalgia n=30 F=22 (73%) INT: 13.6 ±1.8 (12-13) CNT: 12.9 ±2.7 (12-13)	To determine the feasibility of performing a randomized clinical trial to study the effects of an aerobic fitness program; feasibility was defined by program adherence and recruitment ability. Our secondary purpose was to determine the effect of aerobic training on physical fitness (as defined by peak aerobic capacity, muscular power, and metabolic efficiency), Fibromyalgia (FM) symptoms, and overall physical function in children with FM.	Aerobic partial 3/week 40 minutes 12 weeks	Percentage: 64% Overall INT: 67% CNT: 61% "Acceptable": N/R Class register, Self report exercise log (Diary) Objective measure (Heart rate monitor) session attendance Session completed Exercise intensity (Obj)	4.Lack of transportation, 6. lack of time 9. Lack of enjoyment of program	N/R	5. Small instructor/patient ratio (1:4) to ensure adequate attention 7. Video program for home exercises 10. Frequent contact with patients and families through phone to motivate the children and solve potential impediments to participation 10. Children were rewarded with a sticker for each completed exercise session and were able to trade them for small token incentives.
Rathleff *et al.* 2015 (Denmark) Cluster RCT 3/5	Patellofemoral Pain n=121 F=87 (80%) 17.2±1.0 (15-19)	to investigate the effect of exercise therapy as an add-on therapy to patient education compared with education alone on self-reported recovery.	Mixed partial 3/week Supervised: N/R Unsupervised: 15 min Supervised: 12 weeks Unsupervised: 104 weeks	Supervised: 20% Unsupervised: 36% Total: 26% Supervised: ≥80% attendance at prescribed sessions Unsupervised: Exercising on ≥70% of available days Class register, self report exercise log session attendance Exercise frequency	N/R	N/R	10. regular SMS reminders and follow up 5. parent involvement 4. school based exercise sessions 8. multiple choices for timing

Riel *et al.* 2017 (Denmark) RCT 3/5	Patellofemoral Pain n=40 F=35 (88%) INT: 16.9±1.5 (N/R) CNT: 16.5±1.5 (N/R)	to investigate if real-time feedback on contraction time during exercises would improve the ability to perform the exercises with the prescribed contraction time per repetition compared with no feedback on contraction time among adolescents with PFP during a 6-wk intervention.	Strength partial 3/week N/R 6 weeks	Feedback (INT): 35.4% of prescribed total contraction time No Feedback (CNT) 20.3% of prescribed total contraction time N/R Objective measure (App to measure total contraction time, divided by total prescribed time) Exercise time	9. Pain during exercise	10. feedback on contraction time	1. Participants told that compliance to exercises was important and would improve their odds of recovery
Zapata *et al.* 2015 (USA) RCT 3/5	Adolescent Idiopathic Scoliosis n=45 F=29 (85%) 14.9±N/R (N/R)	to investigatewhether 8 weeks of weekly supervised spinal stabilization exercises would reduce pain intensity, disability and functional limitations and improve back muscle endurance in patients with AIS and LBP, as compared with 8 weeks of an unsupervised home exercise program (HEP).	Mixed Partial 3/week 25 minutes 8 weeks	INT: 95% CNT: 67% N/R Self report exercise logs, Interview (verbal report) Session Completion	N/R	6. Decreasing HEP to as few exercises as possible seemed to promote compliance 10. Regular contact (supervised session) promoted improved compliance	9. The exercises were designed to be challenging, fun, and recognizable to promote motivation and adherence to the treatment regimen 7. DVD of exercises was provided to participants
Habers *et al.* 2016 (Netherlands) RCT 2/5	Juvenile Dermatomyositis n=26 F=16 (62%) INT: Median=11.6 (8.3-17.5) CNT: Median=12.6 (8.7-17.6)	to study the feasibility, safety and efficacy of an individually tailored 12-week home-based exercise training programme in the largest group of patients with JDM studied to date.	Mixed partial 2.5/week 40-60 minutes 12 weeks	Percentage: 94% Self report exercise log Objective measure (Heart Rate monitor) Session complete Exercise intensity (Obj measure)	6. Other sport activities 6. holiday 2. fatigue 2. illness 2. transient physical complaints 4. Home based exercise	4. Home based exercise	8. Individually tailored program 4. Provided equipment 7. Provided detail description of program
Eng *et al.* 1993 (Canada) RCT 1/5	Patellofemoral Pain n=20 F=20 (100%) 14.8±1.2 (13-17)	to evaluate the effectiveness of an 8-week program of foot orthotics combined with exercise in adolescent female patients with diagnosed bilateral PFPS	Mixed none 14/week N/R 8 weeks	N/R Reported performance of exercises the previous day on 3 random phone calls. Interview (Self report on phone call) Session completed	N/R	N/R	10. Regular check in phone calls
Quantitative Non-Randomized							
Rivett *et al.* 2014 (South Africa) Pre-experimental 3/5	Idiopathic Scoliosis n=51 N/R N/R	to determine the effect of compliance to the Rigo System Cheneau (RSC) brace and a specific exercise programme on IS curvature; and to compare the quality of life (QoL) and psychological traits of compliant and non- compliant subjects.	Mixed none 5/week 25 minutes >52 weeks	Compliant (INT): 78.4% Non Compliant (CNT) 34.2% Exercising ≥3 days/week Self report exercise log (diary) Exercise frequency	3. Personality traits: emotional instability and low ego strength	3. Personality traits: emotional stability, control, and high ego strength 5. Psychological Support	7.. Written explanations and pictures provided 10. Diary validated with parents 1. Discussion of consequences of non-compliance

Hedayati *et al.* 2018 (Iran) Quasi-experimental 3/5	Idiopathic scoliosis n=34 F=30 (100%) 13.17±N/R (8-17)	to evaluate the impact of group exercise and adjustment of the in-brace force at shorter intervals (twice per week) on Cobb angle and QoL compared with independent exercise and routine brace adjustment (every 3 mos) in patients with idiopathic scoliosis	N/R full N/R 120 minutes 11 weeks	N/R Missing >2 supervised sessions, exercising <6 hr/week at home (total not given so cannot calculate percentages) N/R N/R	N/R	8. Matching patients to preferred treatment (patient choice) 5. Social communication (group better than individual)	N/R
Rathleff *et al.* 2018 (Denmark) Pre-experimental 3/5	Patellofemoral Pain n=20 F=16(80%) 14.6±1.1 (N/R)	to explore adherence to exercise therapy and to use the patient reported outcomes to inform a sample size calculation for a definitive trial.	Mixed Partial 7/week Supervised = N/R Unsupervised= 15 minutes 13 weeks	Supervised: 41% Unsupervised: 50% 80% Supervised= Class roster Unsupervised= self report exercise log (training log) Session attendance, Session Completed	N/R	Home ex preferred over Clinic ex due to: 6. less time intensive nature 4. lower cost 4. ease of implementation 8. Patient choice	5. Parent involvement 10. Regular communication through phone or email 10. Reminder texts day before
Feldman *et al.* 2007 (Canada) Prospective Cohort Study 2/5	Juvenile Idiopathic Arthritis n=175 F=120 (69%) 10.2±4.4 (2-18)	to determine whether adherence to treatment in children with JIA was associated with better clinical, functional, and quality of life outcomes.	N/R N/R N/R N/R N/R	54.2% - 64.1% N/R existing measurement scale (PARQ) Behavioural Component	9. Pain during exercise	2.. younger age, 8. child involvement in responsibility for treatment, and 1. higher perceived helpfulness of the treatment	N/R
April *et al.* 2006 (Canada) Cross-sectional Study 1/5	Juvenile Idiopathic Arthritis n=72 F=41 (82%) 12.67±2.68 (8-18)	to determine the level of agreement between children with JIA and their parents regarding the child's adherence to treatment, for both medication and exercises, and to explore whether factors such as age, disease duration, and disease severity are associated with this agreement	N/R N/R N/R N/R N/R	61.2% (children), 57.4% (parents) N/R Existing measurement scale (PARQ and CARQ) Behavioural Component	1. Negative feelings towards exercises	5. Performing exercises with parents	N/A
Rathleff *et al.* 2016 (Denmark) Pre-experimental 1/5	Patellofemoral Pain n=20 F=18 (90%) 17±N/R (15-19)	to determine if it is feasible to use the exercise monitoring system connected to a tablet device to measure exercise adherence and dosage among adolescents with patellofemoral pain.	Strength partial 3/week N/R 6 weeks	15% of total time under tension prescribed N/R Objective measure (App data) Self report exercise log (Diary) Exercise frequency	10. Adequate feedback during supervised sessions	N/R	1. Participants told that compliance to exercises was important and would improve their odds of recovery
Marais *et al.* 2011 (South Africa) Pre-experimental 1/5	Subtalar overpronation n=20 N/R N/R	to investigate the hypothesis that strengthening the tibialis posterior muscle will decrease subtalar overpronation angles.	Strength partial 5/week 20-30 minutes 4 weeks	N/R N/R Class register, self-report exercise logs session attendance, Session completion	6. school obligations, recreational activities 11. personal priorities	N/R	N/R

Kwan *et al.* 2017 (Hong Kong) Prospective historical cohort matched study 1/5	Adolescent Idiopathic Scoliosis n=48 F=38 (79%) INT: 12.3±1.4 (10-14) CNT: 11.8±1.1 (10-14)	to assess prospectively the effect of Schroth exercise on curve progression, appearance, and QOL in Adolescent Idiopathic Scoliosis (AIS) patients with high-risk curves during bracing.	Mixed Partial 7.5/week Supervised: 60 minutes Unsupervised: N/R Supervised: 8 weeks Unsupervised: N/R	54% of exp group were found to be compliant with exercise > 80% of attendance of therapy sessions and completion of the prescribed home exercise program at least five out of 7 days per week. class register, self-report exercise logs (monitored) session attendance, Sessions completed	N/R	N/R	6. Reduced number and length of sessions from previous protocols to find "compromise between maintaining adequate supervision and minimalizing disruption to the patients' and families' lives"
Carman *et al.* 1985 (USA) Quasi-experimental 1/5	Idiopathic Scoliosis n=45 F=45 (100%) INT: 13.42±N/R (12.08-15.33) CNT: 12.58±N/R (10.5-13.92)	to evaluate prospectively whether a closely monitored physical therapy exercise program, performed in association with Milwaukee brace wear for treatment of scoliosis, had any effect on outcome.	Motor Control none 2/week 120-30 minutes N/R	N/R Rated "good" or "excellent" exercise frequency and quality by physiotherapist at 3-4 month check up visit using arbitrary scale HCP interview (Arbitrary scoring system by physiotherapist for quality and frequency at 3-4 month check up) exercise replication	N/R	N/R	7. Exercise handout
Klepper *et al.* 1999 (USA) Within subject interupted time series 1/5	Juvenile Rheumatoid Arthritis n=29 F=23 (92%) F: 12±N/R (8-17) M: 14±N/R (12-16)	to investigate the effects of an 8-week weight-bearing physical conditioning program on disease signs and symptoms in children with JRA.	Aerobic partial 3/week 60 minutes 8 weeks	N/R "Acceptable": Must have completed at least 18 of 24 available sessions (75%) to be included Class register, Self reported exercise log session attendance sessions completed	9. Pain during exercise	N/R	5. Allowed to have friend exercise with them 10. Parents required to sign activity records 10. Rewards and Incentives for completion 7. Gradual increase in exercise intensity and duration. 7. Careful instruction and monitoring by exercise leaders to ensure safety
Quantitative Descriptive							
Singh-Grewal *et al.* 2006 (Canada) Case series 3/5	Juvenile Idiopathic Arthritis n=9 F=5 (56%) 9.4±N/R (8.9-11.1)	to assess the safety and feasibility of laboratory based exercise testing in children with JIA.	Mixed full 2/week N/R 12 weeks	Percentage:63% "Acceptable": N/R Class register Session attendance	4. Long travel times 6. Family time constraints	9. Enjoyment (pool > gym) 5. Interactions with instructors 10. Small rewards for attendance 6. Scheduling during evening and weekends 6. Duration and frequency	7. Gradual increase in exercise intensity and duration
Wibmer *et al.* 2016 (Austria) Case series 2/5	Adolescent Idiopathic Scoliosis n=8 F=8 (100%) 10.7±N/R (7.8-13.2)	to determine whether an exergame could improve motivation and correct performance of the specific exercises prescribed to treat juvenile and adolescent idiopathic scoliosis.	Mixed none 5/week 30 minutes 24 weeks	32.5% 180 sessions completed Objective measure (Game software: (# of sessions, playing time)) Session completed	9. Bored of exercises if continued for "too long" 7. No progression of difficulty	10. Immediate feedback 9. Playful Diversion 9. Encouragement to improve "score" (within sessions, and between sessions)	N/R

Dovelle *et al.* 1988 (USA) Case study 2/5	Flexor tendon repair of the finger n=1 F=1 (100%) 8	to report on a young girl who received a secondary tendon repair made with a free tendon graft and whose hand was subsequently rehabilitated under the Washington Regimen of early controlled motion.	Mixed none 7/week N/R 12 weeks	N/R N/R interview session completed	N/R	8. Therapists should offer reasonable alternatives, Therapist should remain flexible in approach to patient treatment 5. Establish a positive patient-therapist relationship from the very beginning of therapy	1. Education on importance of exercise to recovery 5. family involvement

Abbreviations: UK, United Kingdom; USA, United States of America; RCT, Randomized Controlled Trial; F, Female; M, Male; INT, Intervention Group; CNT, Control Group; POP, Percentage of Prescribed; N/R, Not Reported; N/A, Not Applicable

** Mixed Method Appraisal Tool. Ratings are from 0 to 5 (with 0 indicating poor quality and 5 indicating excellent quality). Please see Appendix 5 for more information.*

† List of themes: 1. Beliefs; 2. Physical Characteristics; 3. Psychological Characteristics; 4. Physical Environment; 5. Social Environment; 6. Time; 7. Program Details; 8. Engagement; 9. Experience; 10. Reinforcement; 11. Goals

Appendix E: Mixed Methods Appraisal Tool (MMAT) Scoring Guide

Mixed Methods Appraisal Tool (MMAT) Scoring Guide

Replication of instructions from Hong et al[62] with specific customized minimum criteria for this review (underlined), organized by methodological category.

1. Qualitative studies

"Qualitative research is an approach for exploring and understanding the meaning individuals or groups ascribe to a social or human problem" (Creswell, 2013b, p. 3). Common qualitative research approaches include (this list if not exhaustive):

a. **Ethnography**

The aim of the study is to describe and interpret the shared cultural behaviour of a group of individuals.

b. **Phenomenology**

The study focuses on the subjective experiences and interpretations of a phenomenon encountered by individuals.

c. **Narrative research**

The study analyzes life experiences of an individual or a group.

d. **Grounded theory**

Generation of theory from data in the process of conducting research (data collection occurs first).

e. **Case study**

In-depth exploration and/or explanation of issues intrinsic to a particular case. A case can be anything from a decision-making process, to a person, an organization, or a country.

f. **Qualitative description**

There is no specific methodology, but a qualitative data collection and analysis, e.g., in-depth interviews or focus groups, and hybrid thematic analysis (inductive and deductive).

Key references: Creswell (2013a); Sandelowski (2010); Schwandt (2015)

1.1. Is the qualitative approach appropriate to answer the research question?

- Minimum criteria: Describes the approach and rationale for using that method

- Instructions: The qualitative approach used in a study (see non-exhaustive list on the left side of this table) should be appropriate for the research question and problem. For example, the use of a grounded theory approach should address the development of a theory and ethnography should study human cultures and societies. This criterion was considered important to add in the MMAT since there is only one category of criteria for qualitative studies (compared to three for quantitative studies).

1.2. Are the qualitative data collection methods adequate to address the research question?

- Minimum criteria: Refers to the process of how they sampled and collected data: sampling method, description of participants, interviewers, methods/data collection, location
- Instructions: This criterion is related to data collection method, including data sources (e.g., archives, documents), used to address the research question. To judge this criterion, consider whether the method of data collection (e.g., in depth interviews and/or group interviews, and/or observations) and the form of the data (e.g., tape recording, video material, diary, photo, and/or field notes) are adequate. Also, clear justifications are needed when data collection methods are modified during the study.

1.3. Are the findings adequately derived from the data?

- Minimum criteria: Report and describe how data/theory was derived/analyzed (e.g., what approaches they use to get their data, did they use inductive/deductive approaches, thematic analysis, how were theories derived)
- Instructions: This criterion is related to the data analysis used. Several data analysis methods have been developed and their use depends on the research question and qualitative approach. For example, open, axial and selective coding is often associated with grounded theory, and within- and cross-case analysis is often seen in case study.

1.4. Is the interpretation of results sufficiently substantiated by data?

- Minimum criteria: Interpretation/themes supported by data such as quotes. Reports or defines data “saturation” or data/theoretical sufficiency (e.g. Sampling will cease once theoretical or data sufficiency has been achieved. This will be achieved once sufficient data is collected to be able to represent how participants constructed their lives and worlds after this experience and a meaningful reflection of reality is achieved. Or once a

convincing theory or explanation can be confidently verified from the data with no gaps or pitfalls, confidence in theoretical sufficiency has been obtained).

- Instructions: The interpretation of results should be supported by the data collected. For example, the quotes provided to justify the themes should be adequate.

1.5. Is there coherence between qualitative data sources, collection, analysis and interpretation?

- Minimum Criteria: Approach should result in desired outcome of study design (e.g. if doing qualitative description, should not end up in a theory, if used grounded theory, theory should be provided, if looking at a phenomenology, should described the phenomena)
- Instructions: There should be clear links between data sources, collection, analysis and interpretation.

2. Quantitative randomized controlled trials

Randomized controlled clinical trial: A clinical study in which individual participants are allocated to intervention or control groups by randomization (intervention assigned by researchers).

Key references: Higgins and Green (2008); Higgins et al. (2016); Oxford Centre for Evidence-based Medicine (2016); Porta et al. (2014)

2.1. Is randomization appropriately performed?

- Minimum criteria: Describes randomization process AND allocation concealment
- Instructions: In a randomized controlled trial, the allocation of a participant (or a data collection unit, e.g., a school) into the intervention or control group is based solely on chance. Researchers should describe how the randomization schedule was generated. A simple statement such as ‘we randomly allocated’ or ‘using a randomized design’ is insufficient to judge if randomization was appropriately performed. Also, assignment that is predictable such as using odd and even record numbers or dates is not appropriate. At minimum, a simple allocation (or unrestricted allocation) should be performed by following a predetermined plan/sequence. It is usually achieved by referring to a published list of random numbers, or to a list of random assignments generated by a computer. Also, restricted allocation can be performed such as blocked randomization (to ensure particular allocation ratios to the intervention groups), stratified randomization (randomization performed separately within strata), or minimization (to make small groups closely similar

with respect to several characteristics). Another important characteristic to judge if randomization was appropriately performed is allocation concealment that protects assignment sequence until allocation. Researchers and participants should be unaware of the assignment sequence up to the point of allocation. Several strategies can be used to ensure allocation concealment such relying on a central randomization by a third party, or the use of sequentially numbered, opaque, sealed envelopes (Higgins et al., 2016).

2.2. Are the groups comparable at baseline?

- Minimum criteria: Baseline demographics/characteristics are provided (e.g. provides a table with baseline characteristics)
- Instructions: Baseline imbalance between groups suggests that there are problems with the randomization. Indicators from baseline imbalance include: "(1) unusually large differences between intervention group sizes; (2) a substantial excess in statistically significant differences in baseline characteristics than would be expected by chance alone; (3) imbalance in key prognostic factors (or baseline measures of outcome variables) that are unlikely to be due to chance; (4) excessive similarity in baseline characteristics that is not compatible with chance; (5) surprising absence of one or more key characteristics that would be expected to be reported" (Higgins et al., 2016, p. 10).

2.3. Are there complete outcome data?

- Minimum criteria: RCT reports >80% complete data, or <20% drop-out rates
- Instructions: Almost all the participants contributed to almost all measures. There is no absolute and standard cut-off value for acceptable complete outcome data. Agree among your team what is considered complete outcome data in your field and apply this uniformly across all the included studies. For instance, in the literature, acceptable complete data value ranged from 80% (Thomas et al., 2004; Zaza et al., 2000) to 95% (Higgins et al., 2016). Similarly, different acceptable withdrawal/dropouts rates have been suggested: 5% (de Vet et al., 1997; MacLehose et al., 2000), 20% (Sindhu et al., 1997; Van Tulder et al., 2003) and 30% for a follow-up of more than one year (Viswanathan and Berkman, 2012).

2.4. Are outcome assessors blinded to the intervention provided?

- Minimum criteria: Outcome assessors are blinded. Ideally attempts also made to blind the interventionists where possible or reports why they could not blind.

- Instructions: Outcome assessors should be unaware of who is receiving which interventions. The assessors can be the participants if using participant reported outcome (e.g., pain), the intervention provider (e.g., clinical exam), or other persons not involved in the intervention (Higgins et al., 2016).

2.5 Did the participants adhere to the assigned intervention?

- Minimum criteria: 80% of the people follow through on their group allocation. Examples of poor group adherence include >20% crossover from one group/intervention to the other, or reported adherence rates for the intervention are <80%.
- Instructions: To judge this criterion, consider the proportion of participants who continued with their assigned intervention throughout follow-up. “Lack of adherence includes imperfect compliance, cessation of intervention, crossovers to the comparator intervention and switches to another active intervention.” (Higgins et al., 2016, p. 25).

3. Quantitative non-randomized studies

Non-randomized studies are defined as any quantitative studies estimating the effectiveness of an intervention or studying other exposures that do not use randomization to allocate units to comparison groups (Higgins and Green, 2008).

Common designs include (this list if not exhaustive):

a. **Non-randomized controlled trials**

   The intervention is assigned by researchers, but there is no randomization, e.g., a pseudo-randomization. A nonrandom method of allocation is not reliable in producing alone similar groups.

b. **Cohort study**

   Subsets of a defined population are assessed as exposed, not exposed, or exposed at different degrees to factors of interest. Participants are followed over time to determine if an outcome occurs (prospective longitudinal).

c. **Case-control study**

   Cases, e.g., patients, associated with a certain outcome are selected, alongside a corresponding group of controls.

Data is collected on whether cases and controls were exposed to the factor under study (retrospective).

d. Cross-sectional analytic study

At one particular time, the relationship between health-related characteristics (outcome) and other factors (intervention/exposure) is examined. E.g., the frequency of outcomes is compared in different population subgroups according to the presence/absence (or level) of the intervention/exposure.

Key references for non-randomized studies: Higgins and Green (2008); Porta et al. (2014); Sterne et al. (2016); Wells et al. (2000)

3.1. Are the participants representative of the target population?

- Minimum criteria: Inclusion/exclusion criteria reported. Who, where, and how they recruited/sampled is described.
- Instructions: Indicators of representativeness include: clear description of the target population and of the sample (inclusion and exclusion criteria), reasons why certain eligible individuals chose not to participate, and any attempts to achieve a sample of participants that represents the target population.

3.2. Are measurements appropriate regarding both the outcome and intervention (or exposure)?

- Minimum criteria: Reports validity and reliability of outcome of interest, explanation of appropriate measures and justified tools (penalize for lazy reporting)
- Instructions: Indicators of appropriate measurements include: the variables are clearly defined and accurately measured; the measurements are justified and appropriate for answering the research question; the measurements reflect what they are supposed to measure; validated and reliability tested measures of the intervention/exposure and outcome of interest are used, or variables are measured using 'gold standard'.

3.3. Are there complete outcome data?

- Minimum criteria: At least 80% enrolled contributed to outcome of interest

- Instructions: Almost all the participants contributed to almost all measures. There is no absolute and standard cut-off value for acceptable complete outcome data. Agree among your team what is considered complete outcome data in your field (and based on the targeted journal) and apply this uniformly across all the included studies. For example, in the literature, acceptable complete data value ranged from 80% (Thomas et al., 2004; Zaza et al., 2000) to 95% (Higgins et al., 2016). Similarly, different acceptable withdrawal/dropouts rates have been suggested: 5% (de Vet et al., 1997; MacLehose et al., 2000), 20% (Sindhu et al., 1997; Van Tulder et al., 2003) and 30% for follow-up of more than one year (Viswanathan and Berkman, 2012).

3.4. Are the confounders accounted for in the design and analysis?

- Minimum criteria: Controlled cofounders in some way such as study design (matched pairing) or analyses (multi-variable analyses, stratified data)
- Instructions: Confounders are factors that predict both the outcome of interest and the intervention received/exposure at baseline. They can distort the interpretation of findings and need to be considered in the design and analysis of a non-randomized study. Confounding bias is low if there is no confounding expected, or appropriate methods to control for confounders are used (such as stratification, regression, matching, standardization, and inverse probability weighting).

3.5 During the study period, is the intervention administered (or exposure occurred) as intended?

- Minimum criteria: >80% of the people follow through on their intervention or remained in the exposure group. If not reported, it is 0.
- Instructions: For intervention studies, consider whether the participants were treated in a way that is consistent with the planned intervention. Since the intervention is assigned by researchers, consider whether there was a presence of contamination (e.g., the control group may be indirectly exposed to the intervention) or whether unplanned co-interventions were present in one group (Sterne et al., 2016). For observational studies, consider whether changes occurred in the exposure status among the participants. If yes, check if these changes are likely to influence the outcome of interest, were adjusted for, or whether unplanned co-exposures were present in one group (Morgan et al., 2017).

4. Quantitative descriptive studies

Quantitative descriptive studies are “concerned with and designed only to describe the existing distribution of variables without much regard to causal relationships or other hypotheses” (Porta et al., 2014, p. 72). They are used to monitoring the population, planning, and generating hypothesis (Grimes and Schulz, 2002). Common designs include the following single-group studies (this list if not exhaustive):

a. **Incidence or prevalence study without comparison group**

   In a defined population at one particular time, what is happening in a population, e.g., frequencies of factors (importance of problems), is described (portrayed).

b. **Survey**

   “Research method by which information is gathered by asking people questions on a specific topic and the data collection procedure is standardized and well defined.” (Bennett et al., 2011, p. 3).

c. **Case series**

   A collection of individuals with similar characteristics are used to describe an outcome.

d. **Case report**

   An individual or a group with a unique/unusual outcome is described in detail.

   Key references: Critical Appraisal Skills Programme (2017); Draugalis et al. (2008)

4.1. Is the sampling strategy relevant to address the research question?

- Minimum criteria: The source of the sample is relevant to the target population
- Instructions: Sampling strategy refers to the way the sample was selected. There are two main categories of sampling strategies: probability sampling (involve random selection) and non-probability sampling. Depending on the research question, probability sampling might be preferable. Nonprobability sampling does not provide equal chance of being selected. To judge this criterion, consider whether the source of sample is relevant to the target population; a clear justification of the sample frame used is provided; or the sampling procedure is adequate.

4.2. Is the sample representative of the target population?

- Minimum criteria: Inclusion/exclusion criteria are described. Adequate sampling procedure/population
- Instructions: There should be a match between respondents and the target population. Indicators of representativeness include: clear description of the target population and of

the sample (such as respective sizes and inclusion and exclusion criteria), reasons why certain eligible individuals chose not to participate, and any attempts to achieve a sample of participants that represents the target population.

4.3. Are the measurements appropriate?

- Minimum criteria: Explanation of appropriate measures and justified tools, describe the validity and reliability of the tools
- Instructions: Indicators of appropriate measurements include: the variables are clearly defined and accurately measured, the measurements are justified and appropriate for answering the research question; the measurements reflect what they are supposed to measure; validated and reliability tested measures of the outcome of interest are used, variables are measured using 'gold standard', or questionnaires are pre-tested prior to data collection.

4.4. Is the risk of nonresponse bias low?

- Minimum criteria: Include completed data >80%
- Instructions: Nonresponse bias consists of "an error of nonobservation reflecting an unsuccessful attempt to obtain the desired information from an eligible unit." (Federal Committee on Statistical Methodology, 2001, p. 6). To judge this criterion, consider whether the respondents and nonrespondents are different on the variable of interest. This information might not always be reported in a paper. Some indicators of low nonresponse bias can be considered such as a low nonresponse rate, reasons for nonresponse (e.g., noncontacts vs. refusals), and statistical compensation for nonresponse (e.g., imputation). The nonresponse bias is might not be pertinent for case series and case report. This criterion could be adapted. For instance, complete data on the cases might be important to consider in these designs.

4.5. Is the statistical analysis appropriate to answer the research question?

- Minimum criteria: Consistency in the descriptive and inferential statistics (e.g., consistency between using parametric/non-parametric data, matching should be a paired t-tests, or reports met the assumptions for the tests). If no statistical analyses reported, automatic NO

- Instructions: The statistical analyses used should be clearly stated and justified in order to judge if they are appropriate for the design and research question, and if any problems with data analysis limited the interpretation of the results.

5. Mixed methods studies

Mixed methods (MM) research involves combining qualitative (QUAL) and quantitative (QUAN) methods. In this tool, to be considered MM, studies have to meet the following criteria (Creswell and Plano Clark, 2017): (a) at least one QUAL method and one QUAN method are combined; (b) each method is used rigorously in accordance to the generally accepted criteria in the area (or tradition) of research invoked; and (c) the combination of the methods is carried out at the minimum through a MM design (defined *a priori*, or emerging) and the integration of the QUAL and QUAN phases, results, and data. Common designs include (this list if not exhaustive):

a. Convergent design

The QUAL and QUAN components are usually (but not necessarily) concomitant. The purpose is to examine the same phenomenon by interpreting QUAL and QUAN results (bringing data analysis together at the interpretation stage), or by integrating QUAL and QUAN datasets (e.g., data on same cases), or by transforming data (e.g., quantization of qualitative data).

b. Sequential explanatory design

Results of the phase 1 - QUAN component inform the phase 2 – QUAL component. The purpose is to explain QUAN results using QUAL findings. E.g., the QUAN results guide the selection of QUAL data sources and data collection, and the QUAL findings contribute to the interpretation of QUAN results.

c. Sequential exploratory design

Results of the phase 1 - QUAL component inform the phase 2 – QUAN component. The purpose is to explore, develop and test an instrument (or taxonomy), or a conceptual framework (or theoretical model). E.g., the QUAL findings inform the QUAN data collection, and the QUAN results allow a statistical generalization of the QUAL findings.

Key references: Creswell et al. (2011); Creswell and Plano Clark, (2017); O'Cathain (2010)

5.1. Is there an adequate rationale for using a mixed methods design to address the research question?

- Minimum Criteria: Provide a rationale why they used mixed methods (e.g., comprehensive understanding)
- Instructions: The reasons for conducting a mixed methods study should be clearly explained. Several reasons can be invoked such as to enhance or build upon qualitative findings with quantitative results and vice versa; to provide a comprehensive and complete understanding of a phenomenon or to develop and test instruments (Bryman, 2006).

5.2. Are the different components of the study effectively integrated to answer the research question?

- Minimum Criteria: Describes appropriate steps to integrate components into one cohesive product.
- Instructions: Integration is a core component of mixed methods research and is defined as the "explicit interrelating of the quantitative and qualitative component in a mixed methods study" (Plano Clark and Ivankova, 2015, p. 40). Look for information on how qualitative and quantitative phases, results, and data were integrated (Pluye et al., 2018). For instance, how data gathered by both research methods was brought together to form a complete picture (e.g., joint displays) and when integration occurred (e.g., during the data collection-analysis or/and during the interpretation of qualitative and quantitative results).

5.3. Are the outputs of the integration of qualitative and quantitative components adequately interpreted?

- Minimum Criteria: Do the inferences (outputs) make sense given the two components.
- Instructions: This criterion is related to meta-inference, which is defined as the overall interpretations derived from integrating qualitative and quantitative findings (Teddlie and Tashakkori, 2009). Meta-inference occurs during the interpretation of the findings from the integration of the qualitative and quantitative components, and shows the added value of conducting a mixed methods study rather than having two separate studies.

5.4. Are divergences and inconsistencies between quantitative and qualitative results adequately addressed?

- Minimum Criteria: Divergencies are reported AND explained
- Instructions: When integrating the findings from the qualitative and quantitative components, divergences and inconsistencies (also called conflicts, contradictions,

discordances, discrepancies, and dissonances) can be found. It is not sufficient to only report the divergences; they need to be explained. Different strategies to address the divergences have been suggested such as reconciliation, initiation, bracketing and exclusion (Pluye et al., 2009b). Rate this criterion 'Yes' if there is no divergence.

5.5. Do the different components of the study adhere to the quality criteria of each tradition of the methods involved?

- Minimum Criteria: Rate all 3 sections (qualitative, quantitative, mixed methods section, report the lowest score)
- Instructions: The quality of the qualitative and quantitative components should be individually appraised to ensure that no important threats to trustworthiness are present. To appraise 5.5, use criteria for the qualitative component (1.1 to 1.5), and the appropriate criteria for the quantitative component (2.1 to 2.5, or 3.1 to 3.5, or 4.1 to 4.5). The quality of both components should be high for the mixed methods study to be considered of good quality. The premise is that the overall quality of a mixed methods study cannot exceed the quality of its weakest component. For example, if the quantitative component is rated high quality and the qualitative component is rated low quality, the overall rating for this criterion will be of low quality.

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