Adherence in young people living with juvenile arthritis: A systematic review

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Abstract

Objective: Juvenile idiopathic arthritis (JIA) is one of the leading causes of chronic pain in pediatric patients. Treatment regimens, which are critical to symptom management, can be burdensome, involving medication with potentially aversive side effects and exercise that can cause joint pain. Thus, it is important to examine the barriers and facilitators to adherence in JIA. While systematic reviews exist for rheumatic disease in adults, there has not yet been a synthesis of the literature examining adherence in JIA.

Methods: PsychINFO, PubMed and MEDLINE databases were systematically searched to identify qualitative and quantitative empirical studies that investigate adherence for JIA. Keywords included: patient compliance OR adherence OR persistence; youth OR children OR juvenile OR pediatric OR teen OR child OR adolescent; and rheumatoid arthritis OR idiopathic arthritis OR arthritis. Articles were excluded from the review if they involved non-human or adult samples, were non-experimental (e.g., practice recommendations), were not peer-reviewed, or were not written in English. After abstract selection, 32 articles were included in the analyses.

Results: Adherence to exercise regimens was consistently lower than adherence to medication. Researchers relied heavily on self-report of adherence, which suggests a need for additional research with more objective measures of adherence. Across studies, psychological treatment was not included, so adherence to this treatment component in JIA remains understudied.

Conclusions: Results suggest that future research should target devising and evaluating interventions to improve adherence to exercise and perhaps psychological treatment.

Implications for Impact: To facilitate adherence in JIA, behavioral health providers should focus on building a strong therapeutic alliance between provider and child, fostering positive coping skills in parents and children, and monitoring the parent-child relationship.

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Juvenile Idiopathic Arthritis (JIA), formerly known as Juvenile Rheumatoid Arthritis (JRA), is a chronic inflammatory disease that includes several subtypes of chronic arthritis. It is the most common form of arthritis affecting 1 in 1,000 children (age < 16 years) in the United States (Cleveland Clinic, 2017; Hashkes & Laxer, 2005; Mayo Clinic, 2017). Symptoms of JIA during disease flare-ups can be severe and debilitating and typically include inflammation or stiffness in joints, fatigue, and persistent and reoccurring pain, which may lead to problems in daily functioning, mental health, and social interactions (e.g., April et al., 2013; Connelly et al., 2012). JIA has no curative treatment, with symptoms and management typically persisting into adulthood (Cartwright, et al., 2015).

Treatment regimens can vary among patients and between specific JIA subtypes, but typically include medication and exercise components (De Monte et al., 2009). According to the 2021 American College of Rheumatology Guidelines for the Treatment of JIA (Onel et al., 2022), medication may include the use of Nonsteroidal Anti-inflammatory Drugs (NSAIDs), newer biologic therapies, and disease-modifying anti-rheumatic drugs (DMARDs). For others, medication treatment includes Methotrexate, a chemotherapy drug (injected or swallowed) for managing rheumatic pain that is accompanied by potentially severe side effects (e.g., fear of pain from the injection, extreme and oftentimes debilitating nausea, and abdominal pain; Riddle et al., 2006) and immunosuppression (Hashkes & Laxer, 2005). These drug therapies are almost always paired with physical therapy, exercise, and even splinting of joints for pain relief (Jones et al., 2009). While these medications and exercises may ameliorate the pain and inflammation that the child is experiencing in the long-term, aversive side effects or discomfort (e.g., muscle pain from

engaging in exercises; Ramanan et al., 2003) in the short-term may lead to treatment avoidance (Riddle et al., 2006). The aversive aspect of JIA treatment is one of the components that makes the regimen unique. Other treatment regimens for pediatric chronic illness might be similarly demanding and complex with little immediate symptom relief (e.g., diabetes, inflammatory bowel disease); generally though, patients hope for symptom relief upon treatment.

Cognitive behavioral therapy and psychological treatments more broadly have recently been added to treatment regimens for individuals with JIA to offer patients additional, non-pharmacological ways to cope with joint pain and stiffness, or other associated psychological symptoms such as anxiety and depression (Herlin et al., 2015). These psychological treatments also have the potential to provide youth with skills to cope with the negative side effects of medication and exercise, as well as the overall impact that JIA has on their everyday life (e.g., social life, school life) and to learn to adjust their lives to be meaningful despite their diagnosis.

The difficulty and variability in the JIA regimen can make treatment adherence challenging for children, caregivers, and healthcare providers. Because JIA can only be managed and not cured, and given the complexity, demandingness, and highly aversive side effects of treatment regimens, it is not surprising that many patients experience enduring problems with adherence (Kahana et al., 2008). Adherence plays an important role in disease management and morbidity. In fact, adherence in pediatric chronic illness, especially in adolescence, is generally low (e.g., LeLeiko et al., 2013). Despite the importance of adherence, there is no established gold standard for measuring adherence to medication or exercise in JIA (Len et al., 2014). Various self-report measures have been developed specifically for use in measuring adherence in JIA, with one of the most well-validated measures in this area being the *Parent Adherence Report Questionnaire* (De Civita et al., 2005), and its child form, the *Child Adherence Report*

Questionnaire (April et al., 2006). The PARQ has demonstrated moderate to strong construct validity in its assessment of general adherence, adherence to medication, and adherence to exercise. However, as is common with many measures of adherence in pediatric chronic illness (Babyar, 2010, Varni et al., 2015), there are often reporting discrepancies between parent and child, likely due to differing perspectives and involvement in treatment or gaps in knowledge and understanding of the treatment protocol (Len et al., 2014); thus, it can be challenging to achieve absolute accuracy these assessments (April et al., 2006).

The evidence across other pediatric chronic diseases suggests an overall decreasing trend in adherence to medication regimens as children age into adolescence (e.g., LeLeiko et al., 2013), the important role of parents and caregivers in promoting and maintaining adherence (Kroll et al., 1999), and potential barriers to adherence (Lindsay et al., 2011). However, there has not yet been a review that examines adherence in JIA. While a few articles have broadly described and reviewed the nature and challenges of adherence in JIA and other pediatric rheumatic diseases (Rapoff, 1989; Rapoff, 2002; Rapoff, 2006), these articles either do not include a systematic review of the literature (as determined by PRISMA guidelines) or are not specific to JIA. Furthermore, all are dated prior to 2010, thereby not integrating at least 10 years of contemporary research findings (Kroll et al., 1999), including advances in the pharmaceutical treatment of JIA as the use of less aversive and more effective biologic modifiers becomes more common (Bridges et al., 2021). Thus, the aim of this systematic review is to examine existing peer-reviewed literature reporting on adherence in JIA to ascertain adherence rates for different areas of the treatment regimen (e.g., medication, exercise, splinting), identify barriers and facilitators to adherence, and gain understanding of measurement approaches to adherence.

Methods

This study was considered exempt by the primary institution's Institutional Review Board as it did not include any human subjects, only review of published peer-reviewed works. No participants were consented or assented as a part of this project.

Search Strategy

This systematic review followed the guidelines suggested by the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta Analyses). A literature search was conducted using three major databases (PsycINFO, MEDLINE, and PubMed). The purpose of the literature search was to select and evaluate studies that investigated factors associated with adherence, persistence, or compliance for all forms of treatment for JIA. The databases selected are each major databases widely used in systematic reviews (e.g., Gijon-Nogueron et al., 2018; Mehta et al., 2019). Search terms were generated and agreed upon by all authors by identifying keywords of the research question and reviewing keywords used in a previous related systematic review (i.e., Kroll et al., 1999). The following keywords were used as search terms: (patient compliance OR adherence OR persistence) and (youth OR adolescents OR children OR child OR teen OR juvenile OR pediatric) and (rheumatoid arthritis OR idiopathic arthritis OR arthritis). The search was then further limited to articles written in the English language and research conducted on humans.

Study Inclusion Criteria

Articles were included for review if they focused on a pediatric population (0-18 years old) and specifically or separately targeted JIA. The articles had to be original peer-reviewed literature reporting on experimental or clinical trials focused on JIA and adherence to treatment regimens. No treatment regimens were excluded. Treatments included medication,

psychotherapy, exercise, and splinting. Studies had to explicitly measure adherence rates and/or outcomes to be included. There were no restrictions for the date of publication.

Study Exclusion Criteria

Studies were excluded if they were not peer-reviewed (e.g., dissertation, practice recommendation to providers); focused on adult samples; included multiple age groups without results reported specifically for youth; were non-English; did not focus on arthritis or JIA; included multiple disease populations without results separated out for JIA; outcomes were not related to adherence; and/or articles were case studies or did not involve original research.

Study Selection

The PRISMA-P (preferred reporting items for systematic review and meta-analysis protocols) guidelines were followed in the selection of articles for this systematic review (see Figure 1). The search was conducted during September and October 2019, with an updated literature search occurring in September 2021, to acquire any relevant articles published in 2020 and 2021 (5 articles identified and included). The articles were reviewed in two phases. In the first phase, abstracts and titles were reviewed to identify potentially relevant articles to identify studies that examined adherence to treatment regimens in pediatric JIA. In the second phase, of these identified articles, the full-text articles were reviewed by the first and second authors (AKA "coders"). All included and excluded articles were reviewed by coders to come to a final decision on the studies to be included for analysis. If there were any disagreements about the articles meeting requirements, they were discussed among all authors until a final consensus was reached.

Data Extraction

Coders extracted data for this review by hand and stored it in a Microsoft Excel spreadsheet. Extracted data included the total sample size, mean age for the study's sample, measures used, adherence rate, type of adherence measure used, barriers and facilitators to adherence, and methodological strengths and weaknesses. A percentage rate was extracted as a marker of adherence to treatment (e.g., 9/10 days taking medication = 90%). If a study was longitudinal in nature, the reported adherence rates across time were averaged together for a total adherence rate percentage. Barriers and facilitators were extracted if they were specifically noted in the results or discussion of a study.

Data Synthesis

We calculated the average adherence rate and standard deviation for medication, exercise, and splinting across studies, utilizing Microsoft Excel. The same process was utilized to calculate mean age and its standard deviation across studies. To summarize the findings associated with barriers and facilitators, we employed a narrative synthesis approach. This method involves the identification of themes and patterns across studies to summarize the findings, thus going beyond a simple description of the individual studies (Popay et al., 2006). Narrative synthesis guidelines (Popay et al., 2006) guided the presentation and analysis of findings in this systematic review.

Results

Study Selection

The initial electronic search yielded a total of 2,531 results: PsycINFO (N = 70), MEDLINE (N = 706), and PubMed (N = 1,755). After title and abstract review (first phase), 2,474 articles were excluded because they did not meet inclusion criteria (see Figure 1). The most common reason for exclusion was that the article was not peer-reviewed, followed by

studies conducted with adult samples. After duplicates between PsycINFO and MEDLINE were removed, 51 articles were selected for full text review. During the second phase of review, 19 articles were excluded (see Figure 1 for reasons), leaving a final selection of 32 articles for this systematic review (across both review phases). An overview of the included studies as well as data extracted is provided in Table S1.

Study Characteristics

Included studies were published between 1986 and 2021. Ten studies were conducted in the United States, 9 in Canada, 6 in the United Kingdom, 2 in Brazil, and 1 each in Finland, Norway, and Ireland. The study samples comprised a total of 17,992 participants (*M*=562.25 participants, *SD*=2,699.38, Range=8-15,350), with an average mean age of 11.55 years (Range=1-18 years; *SD*=1.71). Participants varied in onset and severity of JIA symptoms across the studies. Studies were diverse in their treatment regimen focus, with some centering on medication (n = 21), exercise (n = 17), or splinting (n = 4). One article also included discussion of diet and comfort measures. None of the articles evaluated adherence to psychological therapies. For the purpose of this study, exercise included structured physical activity (e.g., sports, exercise classes) and physical therapy (e.g., appointments, home exercises). Methodological strengths and weaknesses illustrating the quality of studies, as well as a list of study measures used, are summarized in Table S1. Both coders identified methodological strengths and weaknesses in the design of each study as well as the measures used in the studies (e.g., multimethod approach, subjective vs. objective or direct measurement) and came together to discuss until an agreement was reached. The most common methodological weakness among studies was reliance on self-report measures; indeed, only 7 studies (Adriano et al., 2017; Favier et al., 2018; Hawaa et al., 2015; Litt et al., 1982; Rapoff et al., 2002; Rapoff et al., 2005; Risum

et al., 2018) directly or objectively measured adherence. The most prevalent strengths were the consideration of adherence patterns across time, examination of barriers and facilitators, and use of a variety of study measures.

Adherence Rates

Across the studies that reported an adherence rate, there was substantially higher adherence to medication over exercise or splinting. The mean percentage for medication adherence across all studies was 70.26% (*SD*=18.1). The mean percentage for exercise adherence was 54.99% (*SD*=8.22), while average splinting adherence was 49.25% (*SD*=10.47). However, there was considerable variability in adherence rates across studies: adherence to medication ranged from 30% to 92%, whereas adherence to exercise ranged from 40% to 68%. Generally, studies that used a more objective or direct form of assessing medication adherence (*M*=62.18%, *SD*=15.65, Range=52%-88.3%) found that rates of adherence to medication were lower than those that relied only on self-report (*M*=78.35%, *SD*=17.31, Range=30%-92.2%). Some studies also based medication adherence rates on pharmacy refill data; however, this measurement approach does not account for the possibility that someone can obtain or refill their medication but not take it. Moreover, one study found large discrepancies between parent and child reporting of adherence to medication and exercise (Favier et al., 2018). There is not yet enough evidence from research in JIA to gain consensus as to whether parents or children are more or less accurate in reporting adherence rates; this could be a potential future area of investigation.

Barriers and Facilitators to Adherence

A full list of barriers and facilitators for adherence in JIA are displayed in Table S2. Among the most common barriers to adherence across all studies and all treatment components were ineffective provider and patient communication, exercise being time-consuming, child and

caregiver disagreements over the treatment regimen, the age of the patient (i.e., as children age and seek greater autonomy, poorer adherence arises), and severity of disease (low disease activity is associated with decreased adherence). The most common facilitators to adherence across all regimen components were parental monitoring/supervision of treatment, familial involvement in treatment tasks, recall tools (such as alarms), positive parent coping (e.g., problem-solving, emotion regulation), clear provider communication/explanation of the treatment regimen, greater belief in the effectiveness of treatment, and higher disease severity/fear of disease flare up. Unique to medication, lower side effects acted as a facilitator to treatment adherence.

Discussion

The broad aim of this systematic review was to obtain, summarize, and integrate the existing research investigating adherence in children diagnosed specifically with JIA. Our review demonstrates that adherence to medication in pediatric JIA is consistently better than adherence to exercise. This is not surprising as adherence to exercise recommendations is generally low across pediatric illnesses and even among healthy children (e.g., Holt et al., 2020). However, there was more variability across reports of adherence to medication, possibly due to using different measures and having substantial reliance on self-report, which has been shown to be less accurate than more direct and objective indices (Anghel et al., 2019). Adherence (independent of treatment modality) was generally hindered by familial discord or confusion about the treatment regimen, poor relationship with the child's physician, low disease severity, and frequent and aversive medication side effects. This is interesting, as studies of adherence barriers in other pediatric chronic illnesses identify forgetting medication as the primary barrier to medication adherence (Gutierrez-Colina et al., 2018; Modi & Quittner, 2006). In our review,

adherence was broadly facilitated by a belief that treatment is important and helpful, strong social support, and a greater presence of effective coping behaviors (e.g., emotion regulation skills, problem-solving skills). It appears that for other pediatric chronic illnesses with similarly burdensome regimens, such as Inflammatory Bowel Disease, a belief that treatment is effective is also a powerful predictor of adherence (Greenley et al., 2010).

Aligned with prior work, one major finding of this review was the disproportion between adherence rates to medication compared to those of exercise. Adherence to exercise is much lower than that of medication, despite the long-term benefits of exercise in decreasing symptom severity and pain (Jones et al., 2009). This pattern of difficulty adhering to exercise regimens has persisted over time, as it was identified in research as early as 1985 (i.e., Rapoff et al., 1985). There is often misunderstanding in who assumes responsibility, whether it is the parent or child, for ensuring the child regularly exercises (Greening et al., 2006). Other research suggests there is a parental fear that exercise may hurt the child more, or that the children may be more susceptible to injury due to their disease. Consequently, the parent does not want or allow the child to participate in organized sports or exercise activities (Moola et al., 2008). Additionally, exercise decreases pain in the long run, but may hurt in the short-term for individuals with JIA, and this is challenging for children to comprehend. These potential reasons are congruent with previous work looking across JIA treatment modalities, which identified that children's most common reasons for avoiding exercise therapy included pain, embarrassment, and concern for the future consequences of therapy, among others (Favier et al., 2018). There also may be a gap in provider communication in explaining the benefits of exercise (Blum et al., 2011). Although children living with JIA tend to hold extremities in a position of comfort to avoid pain, exercise greatly reduces joint inflammation and swelling, helps increase range of motion and mobility,

and alleviates pain in the long-term (De Monte et al., 2009). Therefore, regular physical activity or exercise is imperative to reducing the morbidity associated with JIA. To ensure progress is made in this area of adherence, further research and clinical attention to exercise are critically needed to promote adherence and improve health-related quality of life in youth with JIA.

Interestingly, our review highlights the complexities of family or parental support to increase adherence. Some studies found that allowing adolescents greater autonomy over their treatment served as a facilitator for adherence (e.g., Cartwright et al., 2015; Litt et al., 1982), while others found that adherence was superior when parents and children shared responsibility for treatment adherence (e.g., Birt et al., 2014; Hayford & Ross, 1988). While familial involvement may serve as a facilitator for adherence, adolescent autonomy may also be important, creating a difficult balancing act for families. Given that patient/provider communication emerged as a barrier and a facilitator across many studies, and that some studies found that belief in the efficacy of treatment served as a facilitator (e.g., Feldman et al., 2007; Wynn & Eckel, 1986), it is clear that provider communication with families is a critical component to treatment adherence and a possible place for future intervention. Overall, behavioral health providers should work to facilitate strong communication with youth, their families, and their medical team and utilize their expertise to assist families with striking a balance between adolescent autonomy and familial involvement to create an environment where youth can be highly adherent across treatment modalities.

Behavioral health providers are uniquely equipped to employ treatments to facilitate increases in adherence in JIA (and other pediatric chronic illnesses). For example, one existing successful treatment for optimizing adherence includes education around disease and treatment regimen to ensure proper understanding, goal setting and tracking, and development of family

communication and adolescent problem-solving skills (Cushing et al., 2019). The components of this treatment align with many of the barriers and facilitators to adherence identified in this review. In treating JIA, it also is imperative that pediatric psychologists work together to implement evidence-based behavioral techniques to motivate adherence to regimens (e.g., positive reinforcement) and collaborate with healthcare providers to create strong relationships between care teams and families.

It is important to acknowledge that almost all the studies included in the current systematic review relied on self-report measures to examine adherence, which typically are highly subjective to bias (Daniels et al., 2011; Rand, 2000). In our review, results vary depending on the type of adherence assessment used, such that self-report measures produced a wide range of adherence rates, whereas direct measures tended to produce lower rates of adherence and a narrower range across studies (suggesting lower measurement error). Future research in JIA would highly benefit from using more direct measures of adherence or agreeing upon a gold standard of self-report. Other adherence studies of chronic conditions rely on electric monitoring, pharmacy refill data, blood assays for medication, for example, when indexing adherence rates (Lam & Fresco, 2015). Having a reliable, valid, and feasible direct adherence assessment in this population would increase the quality of research and its findings, as well as assist providers in improving adherence rates. Studies and providers should continue to employ already existing objective and direct measures for examining adherence such as electronic monitoring for oral and/or injectable medications (i.e., Medication Event Monitoring Systems; MEMS; Olivieri et al., 1991), pharmacy refill data, and electronic activity monitors that track physical activity.

One of the major limitations of this review was that many of the studies included were conducted prior to 2016; earlier findings on medication adherence in JIA may be somewhat less

applicable, however, given the recent availability of biologics as an alternate form of treatment. Biologic treatment includes a set of medications that block inflammatory cytokines, thereby reducing joint inflammation (Gartlehner et al., 2008). Biologics generally lead to greater treatment efficacy, improved health outcomes, and fewer aversive symptoms relative to more traditional treatments. Thus, the landscape for evaluating and understanding adherence in JIA is ever-changing, suggesting the need to continue evaluating adherence in this population. It is recommended that researchers re-evaluate the nature of adherence to medical regimens in JIA again as new treatments evolve. The articles in this study are, in total, from 7 different countries. Thus, if treatment recommendations are different by country, it may make the results of this review less generalizable. Additionally, articles reporting on other rheumatic disease populations were excluded; however, the barriers and facilitators to adherence remain closely related among these other rheumatic diseases; hence, additional insight into assessing and monitoring adherence in JIA may have been gained by integrating those studies as well (Marengo, et al., 2012).

This systematic review is the first of its kind to examine adherence in JIA, providing researchers and clinicians with a starting point towards improving adherence broadly in this pediatric population, which may offer promise to attenuate disease severity and improve quality of life. Overall, our review provides further evidence that there is a major gap in the literature examining adherence in this population, especially with respect to adherence to psychological interventions, and suggests a critical need for further research to be conducted in this area. Low treatment adherence increases chronic pain and disease morbidity in JIA (Blum et al., 2011). Understanding the barriers, facilitators, and rates of adherence to the treatment regimen is JIA is crucial to improving clinical practice and our understanding of how adherence may be changing over time. In particular, understanding why exercise adherence is so low and finding a gold

standard in assessing adherence to the JIA regimen components would greatly benefit children living with this disease, their caregivers, and their providers. Though psychological interventions are becoming a more standard part of JIA treatment regimens, and hence were included in the search of this review, no articles assessing adherence to psychological interventions were identified, signifying a critical need for research evaluating this form of treatment. Based upon the findings of this review, the need for pediatric psychologists in the treatment of JIA is apparent. Additionally, provider attention should be given to improving exercise adherence, building a strong therapeutic alliance between provider and child, and monitoring the parent-child relationship. In sum, future research is needed using direct adherence measures, large and representative samples, and strong methodological designs to better understand barriers and facilitators to adherence and to subsequently devise paths to improving adherence in youth living with JIA and their families.

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Figure 1. PRISMA Diagram

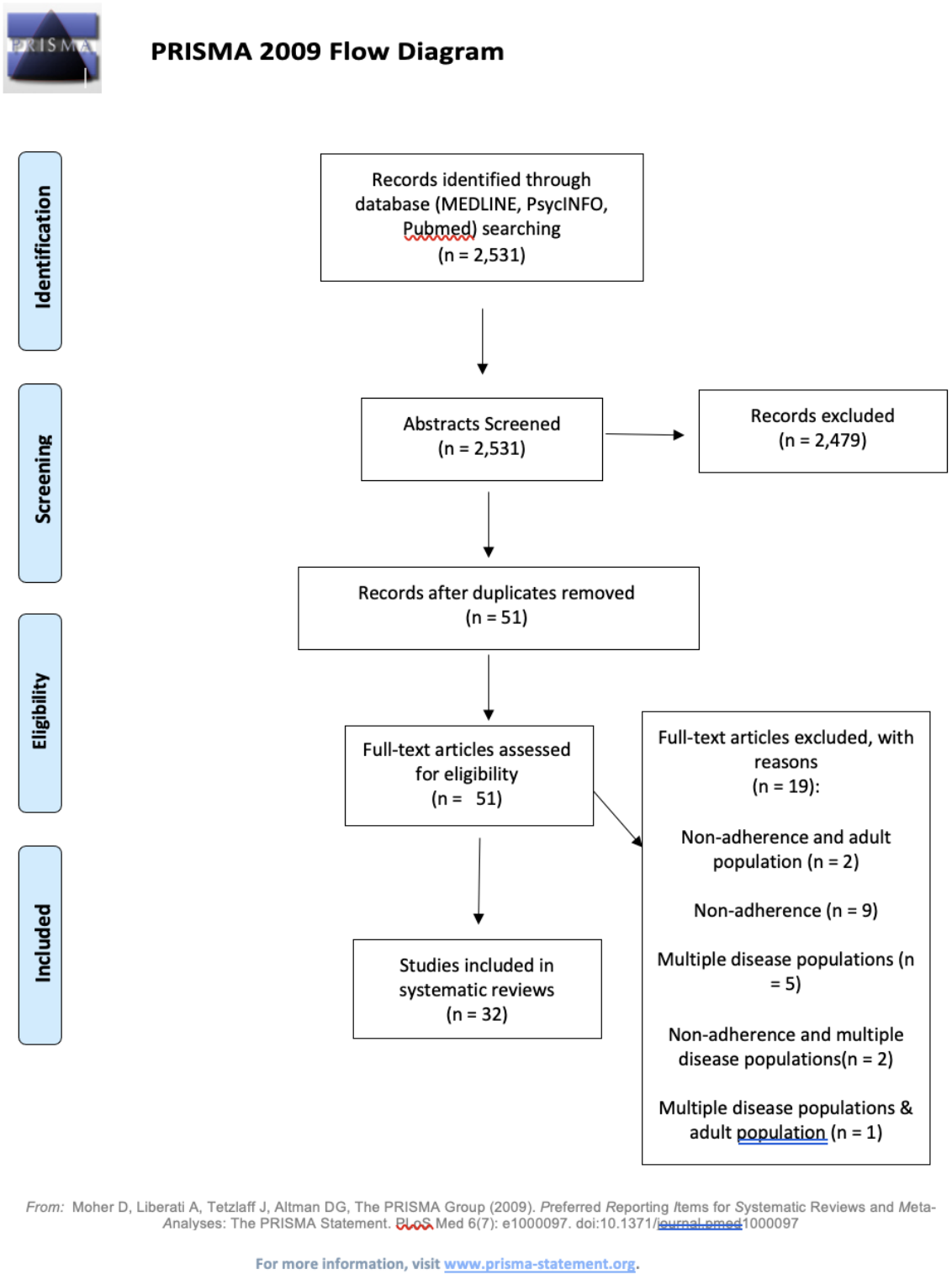

Table 1.

First author, year	n	Age (mean)	Type(s) of adherence measured	Adherence measures used	Adherence rate	Methodological strengths	Methodological weaknesses
Adriano, L. S., 2017	43	11.12	Medication	Analyzed drug dispensing registers of outpatient pharmacies; Parents evaluated for accuracy for each month	53.5% - high adherence; 46.5% -low adherence	Utilized objective & subjective interpretations of adherence; Analyzed differences in medications	Questionnaires used could bias toward higher compliance rates; Medication refill does not mean they will take it
April, K. T., 2006	50	12.67	Medication, Exercise	Parent Adherence Report Questionnaire; (PARQ; De Cevita, Dobkin, Feldman, Karp, & Duffy, 2005); Child Adherence Report Questionnaire (CARQ)	84.9% to medication; 61.2% to exercise	Obtained multiple reports of adherence (parent & child)	Relied on self-report measures only
Birt, L., 2014	28	10.45	Exercise	Qualitative interviews	N/A	Multiple reports were obtained on variables of interest (parent & child)	Parents were present during child interviews, possibly influencing child responses
Brandelli, Y. N., 2019	221	11.10	Medication, Exercise, Splints	PARQ	72% to medication; 68% to exercise, 55% to splints, 69% all treatments combined	Addressed exercise & splinting; Included barriers & facilitators	Relied on self-report measures only; Participants self-selected for participation; Conducted mediation analysis despite cross-sectional data
Britton, C., 1999	46	N/A	Exercise, Splints	Developed a questionnaire	61% to exercise; 39% to splints	Addressed exercise & splinting; Focused on adherence barriers & facilitators	Employed self-report measures only; Did not include medication adherence; Did not use an established adherence questionnaire
Cartwright, T., 2015	10	14.9	N/A_1	Interview	N/A	Utilized interpretive phenomenological analysis	Relied only on self-report measures; Small sample size
Chaney, J. M., 1989	25	12.5	Medication	Self-monitoring diary; Compliance rating question	87.9% to medication	Measured disease activity	Used self-report measures only; Only correlational analyses, thus, no causal inference

De Civita, M., 2007.	175	10.2	Exercise	PARQ	N/A	Focused on barriers & facilitators to adherence to exercise & medication, as well as caregiver recall of treatment protocol	Relied on self-report measures only
Degotardi, P. J., 1999	30	N/A	Medication, Exercise	Quantitative interviews; Personal & disease characteristic questionnaires	40% to medication & exercise; 30% to medication	Employed a quantitative interview method	Employed self-report measures only
El Miedany, Y., 2019	189	12.75	Medication	Medication possession ratio	88.3% to medication in experimental group; 70.5% to medication in control group	More objective measure of adherence than self-report	Using prescription patterns measures possession each month, but not direct adherence
Favier, L. A., 2018	8	12.75	Medication	Medication Event Monitoring Systems (MEMS; Olivieri, Matsui, Hermann, & Koren, 1991); PARQ; CARQ; Developed Barriers to Adherence Tool (BAT)	75.3% to medication	Used both subjective & objective measurements of adherence	Discrepancies existed between parent & child self-report of adherence
Feldman, D. E., 2004	118	10.4	Medication, Exercise, Splints	PARQ	84.9% to medication; 45.9% to exercise; 42% for splints	Employed a variety of different measures; Assessed adherence outcomes	Utilized self-report measures only; Selection bias
Feldman$_1$, D. E., 2007	175	10.2	Medication, Exercise	PARQ	88.4% to medication; 56.6% to exercise	Included a variety of different measures; Measured adherence outcomes	Relied on self-report measures only; Selection bias
Feldman$_2$, D. E., 2007	175	10.2	Medication, Exercise	PARQ	89.1% to medication; 59.2% to exercise	Examined professional perception of adherence; Used a variety of measures	Used self-report measures only; Selection bias; Did not evaluate patterns of adherence
Grande, S. W., 2019	15	12.3		Qualitative interview	N/A	Gathered a depth of patient experience	Did not measure percent adherence

Hawwa, A. F., 2015	49	N/A	Medication	DBS concentrations of MTX active metabolites	57.5% to medication	Directly measured adherence to medication in blood concentrations	Did not evaluate adherence to exercise or other treatment regimens
Hayford, J. R., 1988	93	N/A	Medication, Exercise	Developed compliance & responsibility Likert-type questionnaires	92.2% to medication; 57.1% to exercise	Focused on parent, provider, & child responsibility and how that affected compliance outcomes	Relied on self-report measures only; Did not use established adherence questionnaire
Houghton, K.M., 2018	24	13	Exercise	Weekly activity log, in print or online	47% to exercise	Evaluated multiple outcomes of exercise, including bone outcome & muscle outcome	Focused on adherence to exercise and not other treatment regimens
Kyngas, H., 2002	274	14.6	N/A	Developed 58-item questionnaire	N/A	Focused on predictors of poor adherence; Targeted adolescent difficulties to adherence	Incorporated self-report measures only; Did not use established adherence questionnaire
Litt, I. F., 1982	38	14.2	Medication	Serum salicylate levels	55% to medication	Utilized a more objective form of monitoring adherence	Social desirability may have influenced results of self-esteem measures
Lohse, A., 2021	119	12.4	Medication	CARQ; PARQ; Morisky Questionnaire	75.6% to medication (average of parent and child report)	Utilized two sources for information (parent and child)	Relied upon self-report measures only
Marshall, A., 2019	153 50	10	Medication	Prescription patterns, medication possession ratio	46.6% to biologics; 33.2% to non-biologics	Large sample size	Using prescription patterns measures possession each month, but not direct adherence
Mulligan, K., 2015	116	11.9	Medication	PARQ	74.8% to medication	Examined multiple forms of methotrexate administration (oral & subcutaneous)	Relied on self-report measures only
Pelajo, C. F., 2012	76	10.1	Medication	Adherence was measured by the question: ‘‘How many times did your child miss a dose of prescribed methotrexate in the last 8 weeks?’’ Response options	82% to medication	Assessed barriers & facilitators to adherence rates	Utilized self-report measures only; Did not use established adherence questionnaire; Adherence measured with single question;

				were either 2 times or less, or 3 times or more.			Only studied adherence to medication
Rapoff, M. A., 2005	48	8.6	Medication	MEMS	52% to medication	Used direct measure: electronic monitoring of medication bottle cap openings	Only studied adherence to medication
Rapoff, M. A., 2002	34	N/A	Medication	MEMS	66.9% to medication without study intervention; 77.7% to medication with study intervention	Used direct measure: electronic monitoring of medication bottle cap openings	Only studied adherence to medication
Risum, K., 2018	60	13.5	Exercise	Actigraph GT3X+ accelerometers; Qualitative interview	N/A	Employed objective & subjective measurements of physical activity	Interview information may be biased
Singh-Grewal, D., 2007	80	11.5	Exercise	HAES (Habitual Activity Estimation Scale; Hay, 1997)	56% to experimental treatment (aerobic exercise)	Targeted intensive exercise adherence outcomes	Relied on self-report measures; Focused on effectiveness of study treatment; Did not look at medication adherence
Sims-Gould, J., 2018	17 dyads	N/A	Exercise	Weekly exercise log, on paper or online	46.9% to exercise	Utilized two sources of information for qualitative interviews – parents & children	Relied on self-report measures of adherence
Thompson, S. M., 1995	108	10.04	Medication, Exercise, Splints, Diet, Comfort Measures	Medical chart review	61% to all adherence-facilitating behaviors (medications, splints, exercise, diet, & comfort measures)	Focused on adherence patterns over time	Reviewed medical charts rather than assess adherence directly (e.g., questionnaires)
Toupin-April, K., 2009	182	10	Medication, Exercise	100 mm Visual Analog Scale; PARQ	N/A	Examined both physical exercise & medication	Relied primarily upon self-report measures of

						components of treatment; Relied on multiple reports of adherence (parent, child, rheumatologist)	adherence; Asked about prior 3 months, leading to possible memory bias
Wynn, K. S., 1986	16	9	Exercise	Developed a 56-question questionnaire	N/A	Focused on patient beliefs, adherence barriers & facilitators	Relied on self-report; Used a convenience sample; Used a newly developed, unvalidated questionnaire