Development and Preliminary Reproducibility and Validity of the Parent Adherence Report Questionnaire: A Measure of Adherence in Juvenile Idiopathic Arthritis

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The purpose of this study was to develop and evaluate the psychometric properties of the Parent Adherence Report Questionnaire, a measure of adherence to treatment recommendations in juvenile idiopathic arthritis (JIA). Forty-one primary caregivers of children with JIA (age 1.5 to 18 years) completed a series of measures over a 3-month period. Demographic and disease-related information were obtained at baseline. To measure adherence, caregivers completed bi-weekly diaries on treatment adherence, the general adherence scale (GAS) monthly, and the PARQ at the end of 3 months. The PARQ was reassessed 2 weeks later. Medical charts were reviewed to compare bi-weekly diary reports to actual prescriptions. Intraclass correlation coefficients and kappa coefficients used to assess reproducibility indicated moderate to substantial levels of agreement for PARQ items across time. Construct validity was supported through: (1) significant correlations between the GAS and the PARQ for both exercise and medication; (2) fair agreement between the PARQ-medication and diary reports on medication-related behaviors, and (3) a significant positive correlation between the PARQ-exercise and diary reports on exercise-related behaviors. The study provides preliminary evidence of the reliability and validity of the PARQ.

KEY WORDS: juvenile idiopathic arthritis; adherence, validity; reproducibility; PARQ.

Juvenile Idiopathic arthritis (JIA) is the most common type of chronic arthritis to affect children. By definition, it has its onset before 16 years of age. It has a mean age of onset of 5 years (Cassidy & Petty, 2001) and persists into adulthood in up to 50% of cases (Oen et al., 2002). This entity comprises what has generally been referred to as juvenile rheumatoid arthritis (JRA) in North America and juvenile chronic arthritis (JCA) in Europe. There are seven onset types—the most common of which is oligoarthritis (Petty et al., 1998). Although the precise incidence of JIA is not known, one can infer that it approximates to 50% greater than that of JRA, which varies between 5 to 20 per 100,000 children less than 16 years of age (Bowyer, Roettcher, & the Members of the Pediatric Rheumatology Database Research Group, 1996; Kunnamo, Kallio, & Pelkonen, 1986;

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Malleson, Fung, & Rosenberg, 1996; Symmons et al., 1996). The rationale behind this estimate lies in the inclusion of psoriatic arthritis and enthesitis-related in JIA, which approximates to 50% of JRA.

Children with JIA are prescribed medications to control inflammation, to modify the disease, and to reduce pain. They are also prescribed physical and occupational therapies, which are important to prevent contractures and to maintain joint functioning (Wara & Emery, 1991). Studies involving children with JIA report that adherence to medication is better than to either exercises or the use of splints (Degotardi, Revenson, & Ilowite, 1999; Hayford & Ross, 1988; Rapoff, Lindsley, & Christophersen, 1985). Less than optimal adherence to JIA therapies can lead to missed school days (Sturge, Garralda, Boissin, Doré, & Woo, 1997) as well as compromise functional status (Rapoff, 1999). On a societal level, adherence failures can reduce the cost-effectiveness of the medical care system, negatively impact clinical decision-making, and bias clinical trials of promising therapies (Rapoff, 1999).

By and large, researchers have tended to approach the study of adherence using Haynes' (1979) original definition of *compliance* as the "extent to which a person's behavior coincides with medical or health advice" (pp. 1–2). This brings research to focus on determining a 'match' between patient treatment-related behaviors and recommendations. Pediatric adherence, however, is more encompassing in that it is multidimensional; it involves caregivers in treatment; and both caregivers and children play an active role in determining how the treatment plan is managed (De Civita & Dobkin, 2005).

The term 'multidimensional' refers to the complexity of most treatment plans for chronic illnesses. Interventions often include several components (e.g., medications, exercises, splints), with different behaviors (e.g., taking oral non-steroidal anti-inflammatory medication three times a day; performing range of motion exercises) being executed on a regular basis to maintain optimal health. Children's ability to follow recommendations in terms of frequency, general level of difficulty, and negative reactions (e.g., crying, complaining, refusing) toward treatment may therefore differ depending on the type of therapy. Instruments that assess children's ability in relation to carrying out different treatment-related behaviors provide a broader perspective from which to consider difficulties experienced by children in maintaining optimal functioning.

A second consideration for pediatric adherence is caregiver involvement in treatment. In early childhood, caregivers are completely responsible for ensuring that treatment recommendations are met. With increasing developmental maturity, children with JIA may assume greater responsibility for their treatment (Degotardi et al., 1999; Hayford & Ross, 1988). Research in Type 1 diabetes suggests, however, that possessing adequate knowledge about disease management may not necessarily mean that the child knows how to effectively execute treatment tasks (La Greca, Follansbee, & Skyler, 1990). Caregivers may therefore remain solely responsible for some components of treatment and/or share responsibility. When tasks are shared, there is the possibility of disagreements with regard to who is primarily responsible for a particular component of treatment (Hayford & Ross, 1988). By understanding how treatment tasks are distributed within the family, health care providers may be better positioned to address treatment-related problems.

A third aspect of pediatric adherence relates to the active role caregivers and children play in determining how treatment for JIA is managed. Caregivers, for instance, may choose an alternative course of action, one that is consistent with their beliefs about the efficacy of treatment regimens. Studies involving children with JIA report that caregivers' perception regarding the helpfulness of therapies encourages their commitment to the treatment plan (Schroder, Crabtree, & Lyall-Watson, 2002; Wynn & Eckel, 1986). Health professionals may gain considerable insight into how caregivers feel about their child's treatment plan by inquiring about their perception regarding the helpfulness of different therapies. However, even when caregivers believe in its efficacy, they may still have concerns with treatment side effects (Kroll, Barlow, & Shaw, 1999). Stopping medication when child is feeling worse, for instance, may reflect caregiver concern with the medication regimen. Alternatively, caregivers' approach to managing their child's treatment may be affected by their understanding of instructions (Rapoff, 1999) and whether they felt that concerns related to their child's condition were adequately addressed by health care providers (Barlow, Harrison, & Shaw 1998). Equally important is to understand the role that children play in ensuring the successful management of treatment. For instance, they may choose not to follow recommendations as a means of expressing their independence and autonomy from their family (Kazak, Segal-Andrews, & Johnson, 1995) and/or for

fear of appearing different from their peers (Barlow et al., 1998; Rapoff et al., 1985; Schroder et al., 2002). Health care providers would therefore need to be cognizant that children and caregivers may encounter barriers in implementing the treatment plan.

Health professionals working with patients diagnosed with JIA would benefit from a measure that conceptualizes adherence as being multidimensional, involving caregivers in treatment, and acknowledges the active role caregivers and children play in determining how treatment is managed. This requires assessing for child ability in relation to different treatment components (i.e., frequency, general level of difficulty, and negative reactions toward therapies) and caregiver perception regarding the helpfulness of therapies. It also calls for the assessment of how treatment tasks are distributed within the family. As well, understanding how caregivers and children are managing treatment requires identifying potential barriers to treatment such as medication side-effects and not understanding instructions. In addition, assessing for errors in medication behavior may prevent deterioration in health status. Caregivers and/or children may forget medication instructions, be careless about administration, or discontinue medication when symptoms subside or when side-effects are apparent (Rapoff, 1999).

To our knowledge, there does not exist such a measure of adherence in JIA. The Compliance Questionnaire used in JIA by Rapoff et al. (1985), for example, acknowledges the importance of assessing child ability in relation to different treatment components; albeit, it does not allow health care providers to identify whether caregivers or children are primarily responsible for some components and share responsibility for others. Measures have been developed to assess family division of responsibility for asthma treatment (McQuaid et al., 2001) and for Type 1 diabetes treatment (Anderson, Auslander, Jung, Miller, & Santiago, 1990). Despite the fact that Hayford and Ross (1988) assess for the distribution of treatment responsibility in JIA, they use a single item to determine the frequency of taking medication and performing exercises as prescribed. They do not assess for children's general level of difficulty or negative reactions in response to following treatment as prescribed. Similarly, Wynn and Eckel (1986) simply asked caregivers to rate the frequency of their child's exercise-related behaviors. Likewise, Struge et al. (1997) used a single item to assess children's cooperation with wearing splints, taking medication, and doing exercises. Other

research involving children with JIA has used family interviews to determine children's treatment-related behaviors and the distribution of responsibility for treatment tasks (Degotardi et al., 1999). Qualitative approaches have also provided valuable information about caregiver perception regarding the helpfulness of therapies (Schroder et al., 2002) and potential barriers to implementing treatment (Barlow et al., 1998). Although such research has provided ample information regarding aspects of adherence, it has been gathered in a piece-meal fashion.

The goals of this study were to: (a) describe the Parent Adherence Report Questionnaire (PARQ); and (b) assess its reproducibility and construct validity. The PARQ was designed to gain an understanding of: children's ability in taking medications, doing exercises, and wearing splints; family distribution of treatment responsibilities; caregivers' perception of the helpfulness of therapies; errors in taking medication; and potential barriers to treatment. To develop the PARQ, studies on adherence in general as well as specific to JIA were critically reviewed. Regarding the distribution of treatment tasks, we expected to observe shared responsibility. In support of a multidimensional approach, we expected to find significant differences in child ability scores and caregiver perceived helpfulness across treatment components. Caregivers' perception of helpfulness was expected to be positively associated with better child ability scores. Regarding the PARQ's reproducibility, we hypothesized significant agreement between items over a two-week interval period. This period was considered to be long enough to prevent recall bias and short enough to ensure stability in adherence characteristics. In support of its construct validity, we hypothesized that child ability scores in relation to different treatment components would be positively associated with a general adherence measure and would show significant agreement with estimated adherence rates. Estimated adherence rates reflect self-monitoring of treatment-related behavior in relation to prescriptions as recorded in medical charts, and as such are considered to be a measure of actual performance. Children's level of difficulty, negative reactions, and the frequency of following recommendations are salient features when monitoring behavior. Therefore, comparing the PARQ child ability scores to estimated adherence rates would provide a more rigorous evaluation of its construct validity.

METHOD

Participants

Primary caregivers of children with JIA were recruited from the JIA clinic at the Montreal Children's Hospital (MCH) between March 2001 and November 2001. A total of 188 visited the clinic during this period. To be eligible for the study, caregivers had to be fluent in either English or French, their child had active disease (i.e., not in remission) and was prescribed medication for JIA. Not all children were prescribed exercises and/or splints. One hundred and six (56.4%) caregivers fulfilled study criteria. From this pool of eligible subjects, 73 (68.9%) were approached. Twelve caregivers declined after being informed about the study, leaving a sample of 61. Reasons for refusal included lack of time or disinterest in the study.

Measures

Parent Adherence Report Questionnaire (PARQ)

A review of the literature on adherence and several consultations with a team of health professionals (pediatric rheumatologists, physical therapists, occupational therapists, pediatric nurses, and a health psychologist) led to the creation of an initial pool of items, which formed the basis of the PARQ. The team was consulted for a second time to provide feedback about face validity, content (i.e., accuracy and clarity of the questions), and application (i.e., time required to complete). Next, PARQ was pretested for comprehensibility and ease of use in five English-speaking caregivers of children attending the JIA clinic at the MCH. Minor changes were made and it was translated into French by a bilingual professional translator.

The PARQ consists of three sections. Instructions are prefaced with 'in the past 3 months' to indicate time elapsed since the baseline visit when treatment was prescribed. Section 1 identifies who is primarily responsible for different treatment components—namely, taking medication, doing exercises, and wearing splints: (1) mostly mother's responsibility; (2) mostly father's responsibility; (3) mostly child's responsibility; and (4) mostly someone else's responsibility. This question was cited in a study by Hayford and Ross (1988) on medical compliance in JIA.

Section 2 measures child ability in relation to each treatment component. Caregivers indicate on a 100-mm horizontal visual analog scale (VAS): (1) child's general level of difficulty in following treatment recommendations; (2) how often child follows treatment recommendations as prescribed by the health care provider; and (3) how often child shows any negative reactions such as crying, complaining, or refusing to cooperate in response to following recommendations. The endpoint anchors of the VAS for item 1 are '0 = very easy' and '100 = very hard'; and for items 2 and 3 they were '0 = never' and '100 = always'. Responses to items 1 and 3 are reversed so that higher scores indicate fewer difficulties and fewer negative reactions, respectively. An overall child ability score can then be computed separately for medication (PARQ-CA-medication) and exercise (PARQ-CA-exercise) by averaging responses to the three items respectively. Pearson's r among the child ability items ranged from 0.45 to 0.86 for medication and 0.62 to 0.77 for exercise. Higher scores indicate better overall child ability in relation to following treatment recommendations. The next four questions in this section ask whether they or their child made errors in medication behavior using a yes/no format: (1) ever forget to take medicine; (2) being careless at times about taking medicine; (3) when feeling better sometimes stopped taking medicine; and (4) when feeling worse when taking medicine, sometimes stopped taking it. Positive responses are summed, with total scores ranging from 0 (no errors) to 4. These questions were adapted from Morisky, Green, and Levine's (1986) self-report measure of adherence to medication regimens for adult patients.

Section 3 asks about caregiver perception regarding the helpfulness of each treatment component (e.g., medication, exercises, and splints) using a 100-mm horizontal VAS. Endpoint anchors are '0 = not helpful' and '100 = very helpful'. Caregivers are also asked about the therapies they most preferred and why. Answers to these questions have yet to be coded and will not be presented in this study. The final question in this section inquires about potential barriers to treatment. Response categories include: A change in child's rheumatologist; medication side effects; treatment recommendations too hard to follow; follow-up appointments too infrequent; poor coordination with other services; too long a wait at each appointment; medical explanations about condition or treatment unclear; explanation for therapies unclear; and other barriers.

Demographics Questionnaire

The demographic questionnaire included questions on child's age, cultural background, and living arrangements. The caregiver was also asked questions about annual family income, employment status, and the highest level of schooling completed by both caregivers. In this study, we focused on mother characteristics.

Physician Form: Disease Characteristics

The JIA clinic at the MCH uses a standardized system of documentation of patient status at each visit in terms of disease severity. Disease severity was determined by the sum of joint severity score, which is computed by summing three subscales: (1) effusion, scored on a scale of 0 to 3; (2) tenderness, scored on a scale of 0 to 3; and (3) loss of range of motion, scored on a scale of 0 to 4, for all joints, in accordance with the Pediatric Rheumatology Collaborative Study Group (Giannini & Brewer, 1982). Patients are classified according to the International League Against Rheumatism by practitioners experienced in using this classification system. For the purpose of this study, disease severity at baseline was used. Disease duration and age at disease onset were recorded in years.

General Adherence to the Treatment Plan

The General Adherence Scale (GAS; Sherbourne Hays, Ordway, DiMatteo, & Kravitz, 1992), modified by the research team, was used to measure children's general adherence to their treatment plan. Caregivers completed the GAS at the end of each month for a period of 3 months. They were asked to respond to five questions about their child's general level of difficulty and frequency in following treatment recommendations, regardless of the type of intervention, using a Likert-type scale with 1 indicating 'none of the time' and 6 indicating 'all of the time'. Items included: (1) your child had a hard time doing what the health care providers suggested he/she do; (2) your child followed his/her health care providers' suggestions exactly; (3) your child was unable to do what was necessary to follow his/her health care providers' treatment plans; (4) your child found it easy to do the things his/her health

care providers suggested he/she do; and (5) generally speaking, how often was your child able to do what his/her health care providers told him/her? The instruction to this questionnaire was prefaced with 'during the past 4 weeks' to indicate time elapsed since the last month. Monthly total scores were averaged to create a global score, capturing general adherence to the treatment plan. In their study, Sherbourne et al. reported an internal consistency reliability of .78, with a 2-year stability of $r = .41$. In this study, internal reliability coefficients for month 1, 2, and 3 were 0.77, 0.79, and 0.69, respectively.

Estimated Adherence Rates

Caregivers were asked to complete 2 one-week diaries per month for each of the 3 months, and for each treatment component (a total of 42 days per treatment component). For medication, they were asked to indicate the type of medication prescribed and the frequency per day. For prescribed exercises, they were asked to describe the exercises recommended, the number of repetitions, and sessions per day. For splints, they were asked to indicate the type(s) of splints and hours worn per day. Diaries were compared to actual prescriptions to derive estimates of adherence. Estimated adherence rates were operationalized as the ratio of reported frequency of following treatment prescriptions as recommended [i.e., taking medication(s), doing exercise(s), and wearing splint(s)] over the 3-month period to the frequency of actual prescribed treatment, as indicated in the patient's medical chart. For example, a child prescribed methotrexate one time per week would need to report six occasions in which the drug was taken over the 3-month study period (i.e., 2 reports per month) to have an adherence rate of 100%. If, however, the caregiver reported that the child took methotrexate only 5 times during the study period, his/her adherence would be 83%. Separate estimates of adherence to exercise and splints regimens were computed. Changes in treatment recommendations during the 3-month study period were recorded and taken into account when calculating estimated adherence rates.

Procedure

This study was part of a larger research program, which aims to identify determinants of adherence

and quality of life in children with JIA. Caregivers who met study criteria were approached during regular clinic visits by a project coordinator. At study entry, caregivers completed the Demographics Questionnaire. Other measures on child quality of life, caregiver distress, coping behaviors, use of alternative therapies, and perceived economic hardship were also completed, but not considered in this study. During a 3-month period, they completed bi-weekly diaries pertaining to specific treatment recommendations and the GAS at the end of each month. At the end of month 3, caregivers completed the PARQ. Two weeks later, the PARQ was completed again. Caregivers were instructed to return all questionnaires using pre-addressed stamped envelopes. Health care providers examined children at baseline and at the 3-month period. Medical charts were reviewed to determine the accuracy of diary reports of treatment-related behaviors. The Institutional Review Board of MCH approved this study and all participants provided informed consent. The entire pediatric rheumatology team participated in the study.

Statistical Analysis

Descriptive statistics, including means, medians, standard deviations, and ranges were computed for all study variables. Paired t-tests were conducted to compare the PARQ child ability and helpfulness items between treatment components. Pearson correlation coefficients were computed to determine the degree of association among the PARQ child ability and perceived helpfulness items across treatment components.

Reproducibility of the PARQ's VAS scores was analyzed using the intraclass correlation coefficient (ICC), which is the proportion of the between subject variance to the total variance. We considered ICC values greater than .70 as indicative of substantial concordance over time and values less than .70 as indicative of moderate to poor concordance (Portney & Watkins, 1993). The kappa statistic was used to measure agreement for categorical data (i.e., locus of responsibility; yes/no items on medication-taking). *A priori*, we considered kappa values >.81 as representing almost perfect agreement; values from .61 to .80 as substantial agreement; values from .41 to .60 as moderate agreement; values of .21 to .40 as fair; and below .21 as indicative of slight agreement (Landis & Koch, 1977).

Construct validity was determined through a series of analyses. First, Pearson correlation coefficients were computed to determine the degree of associations between GAS and PARQ-CA scores. Second, correlation coefficients and kappa coefficients were computed to examine consistency and agreement, respectively, between overall child ability scores and estimated adherence rates. Separate analyses were conducted for PARQ-CA-medication and PARQ-CA-exercise and associated estimated adherence rates. In computing kappa, the distribution of scores on both measures was split at the clinical value of <80%. Rapoff (1999) suggests a minimum criterion of less than 80% for classifying patients as nonadherent to prescribed regimens. The distribution of scores for both estimated medication adherence and PARQ-CA-medication were significantly truncated, thereby violating the assumption of bivariate normality required for computing correlations. Therefore, we examined only the concordance between these two measures. Only five patients were prescribed splints as part of their treatment analysis. Analysis therefore focused only on the medication and exercise regimens.

RESULTS

Description of Study Sample

Of the 61 caregivers enrolled in the study, 16 (26.2%) withdrew before the 3-month study period, leaving an initial sample of 45. The major reason for withdrawal was lack of time for completing the study questionnaires. Of the 45 caregivers who completed the study, one did not complete the Demographics Questionnaire and three did not complete at least three bi-weekly diaries on medication (a total of 21 days), leaving 41 (91.1%) subjects for the analysis. Analyses involving exercise regimens were conducted with a reduced sample (n's ranging from 19 to 35); albeit those prescribed exercises had to have diary reports on at least 21 days over the 3-month study period.

No significant differences were found on child characteristics (e.g., gender and age), maternal education, and disease characteristics for these 41 versus the 20 who either withdrew or were dropped from the analysis due to missing data. A significant mean difference was found for maternal age, with mothers retained in the analysis being older ($p = .0009$). Among the 41 respondents, 58.54% were mothers with a mean age of 40.85 ($SD = 5.78$; range $= 30$ to 60 years).

Table I. Demographic, Disease, and Treatment Characteristics of Children Retained in the Study ($N = 41$)

	N (%) or Mean ± SD (Min, Max)
Demographics:	
Child gender (Girls %)	31 (75.6)
Child age (years)	10.4 ± 4.2 (1.5, 18)
Cultural background (%)[a]	
French Canadian	17 (42.5)
English Canadian	6 (15.0)
European	6 (15.0)
Mexican/Central or South America	1 (2.5)
Asia	1 (2.5)
Other	9 (22.5)
Family configuration (%)	
Two-parents	31 (75.6)
Mother-only	6 (14.6)
Shared custody	1 (2.4)
Other	3 (7.3)
Disease:	
Disease type (%)	
Oligoarthritis	14 (34.1)
Polyarthritis-RF negative	11 (26.8)
Polyarthritis-RF positive	1 (2.4)
Systemic onset	3 (7.3)
Psoriatic arthritis	3 (7.3)
Enthesitis-related arthritis	8 (19.5)
Other[b]	1 (2.4)
Age at disease onset (years)	6.1 ± 4.4 (0.7,15.6)
Duration of disease (years)	4.4 ± 3.9 (0.02,15.1)
Sum of joint severity	8.7 ± 15.5 (0,76)
Treatment components	
Medication (%)	18 (43.9)
Medication and exercise (%)	18 (43.9)
Medication, exercise, and splints (%)	5 (12.2)

Note. [a]One subject refused to provide data; [b]unclassified JIA.

Twenty-five respondents (60.9%) indicated English as their mother tongue. Approximately, 60.9% ($n = 25$) of mothers had more than a high school degree and 46.3% ($n = 19$) were employed full time. Median family income was 70,000 (range =14,999 to 95,000).

Table I presents the demographic, disease, and treatment characteristics of the 41 children. Most of the children (57.5%) were of Canadian background and 75.6% were girls. Median age was 11.4 years. The majority resided in two-parent families. Most were diagnosed with oligoarthritis (34.1%). Median age at disease onset was 4.7 years. Median disease duration was 3.5 years. Wide variability was observed for disease severity, as measured by the sum of joint severity count, with a mean of 8.7. This score is indicative of mild severity. Over 40% of children were prescribed only medications. An equal proportion of children following a medication regimen also had exercises as part of their treatment. Only five children were prescribed

medications, exercises, as well as instructed to wear splints.

Description of the PARQ

Table II presents descriptive statistics for each item on the PARQ. Original categories for the locus of responsibility for medication and exercise regimens were collapsed into: (1) caregivers; (2) child only; and (3) caregiver and child. In general, either the caregiver or the child was primarily responsible for ensuring that treatment recommendations were followed. Shared responsibility was more common for the exercise regimen as opposed to the medication regimen. Results from a series of paired *t*-tests revealed significant differences for child ability and caregiver perceived helpfulness between treatment components. According to caregivers, children showed significantly less difficulties ($t = -5.30$, $p < .0001$) and negative reactions ($t = -2.35$, $p = .025$) as well as a greater tendency to

Table II. Characteristics of the PARQ

Questionnaire items	N (%) or Mean ± SD (Min, Max)
Section 1: Responsibility for Treatment (%)	
Medication regimen	
Caregivers (mother, father, both caregivers)	26 (63.4)
Child only	13 (31.7)
Caregiver and child	2 (4.9)
Exercise regimen ($n = 36$)	
Caregivers	18 (50)
Child only	14 (38.9)
Caregiver and child	4 (11.1)
Section 2: Child ability	
General level of difficulty in following treatment[b]	
Medication	86.3 ± 22.0 (21.8,100)
Exercise ($n = 35$)	59.3 ± 29.2 (0,100)
Frequency of following treatment	
Medication	90.2 ± 17.4 (11,100)
Exercise ($n = 33$)	58.3 ± 32.7 (0,100)
Negative reactions in response to following treatment[b]	
Medication	87.9 ± 19.9 (20.8,100)
Exercise ($n = 35$)	73.8 ± 32.8 (0,100)
Total PARQ Score: Child ability	
Medication	88.2 ± 17.0 (43.2,100)
Exercise ($n = 35$)	64.0 ± 27.9 (0,100)
Made errors in taking medication (%)	25 (60.98)
Section 3: Helpfulness of therapies	
Medication	88.3 ± 18.3 (22.8,100)
Exercise ($n = 33$)	75.6 ± 28.5 (0,100)
Experienced barriers to treatment[c] (%)	17 (42.5)

Note. [b]Higher scores indicate less difficulties and negative reactions. [c]$n = 40$ patients.

follow medication recommendations as prescribed ($t = 5.52$, $p < .0001$) compared to exercise regimens. Overall child ability (PARQ-CA) scores were significantly better for medication than exercise ($t = 4.82$, $p < .0001$). No statistically significant difference was found between caregivers' perceived helpfulness of medication versus exercise treatment ($t = 1.77$, $p = .087$). None of the child ability items in relation to medication regimen correlated significantly with caregiver perceived helpfulness of that regimen. Perceived helpfulness of exercise regimen was positively and moderately correlated with how often child followed exercise recommendations [$r(32) = .49$, $p = .0041$], applying a Bonferroni correction ($.05/6 = .0083$).

Regarding errors in medication-taking behaviors, 22 (55%) caregivers reported that they or their child forgot to take medication. Seven (17.1%) indicated being careless at times and five (12.2%) reported stopping medication when child was feeling better. No caregiver reported stopping medication when the child was feeling worse. Seven (17.1%) reported making two or more errors when taking medication. Caregivers also reported barriers to their child's treatment. Most frequently reported barrier was medication side-effects ($n = 8$, 20%), followed by waiting too long at each appointment ($n = 7$, 17.5%). Less noted barriers included: a change in child's rheumatologist ($n = 1$, 2.5%); treatment recommendations hard to follow ($n = 3$, 7.5%); and poor coordination among services ($n = 2$, 5%). The 'other category' elicited two more barriers-namely, explaining child's situation to new medical residents ($n = 1$, 2.6%) and having no set date for surgery ($n = 1$, 2.6%). Three caregivers (7.5%) reported more than one barrier to their child's treatment.

Reproducibility and Validity of the PARQ

Estimated medication adherence rates ranged from 16.7 to 100%, with a mean of 86.4 ($SD = 18.3$). Its distribution was negatively skewed, with the median being 96.6%. Estimated exercise adherence rates ($n = 21$) ranged from 0 to 100, with a mean of 52.3 ($SD = 35.9$). This distribution was slightly skewed to the left, with a median of 66.7. The mean level of general adherence as measured by the GAQ for 32 children was 5.1 ($SD = .72$, range = 2.9 to 6). Its distribution was skewed to the left, with a median of 5.2.

Reproducibility

ICCs used to assess reproducibility of the child ability scores related to medication regimen between the two periods of assessment indicated moderate concordance for level of difficulty (ICC = .62) and frequency of following treatment (ICC = .60). Scores on negative reactions (ICC = .38) over time indicated poor concordance. Intraclass coefficient for helpfulness of medication regimen represented moderate concordance (ICC = .55). ICCs for child ability items related to exercise were higher for frequency of following exercise regimen (ICC = .88) and level of difficulty with treatment (ICC = .82) compared with negative reactions (ICC = .53). Intraclass coefficient for helpfulness of exercise regimen represented substantial concordance (ICC = .79).

Kappa coefficients for the locus of responsibility for regimens represented substantial agreement for medication [$\kappa(36) = .72$, 95% Confidence Intervals (CI): .50, .94] and moderate agreement for exercise [$\kappa(31) = .42$, 95% CI: .15, .69], beyond chance. Kappa coefficients for the four items on errors in taking medication indicated substantial levels of agreement for forgetting [$\kappa(35) = .65$, 95% CI: .40, .90)] and for being careless [$\kappa(36) = .62$, 95% CI: .32, .92]. The obtained kappa value representing agreement on stopping medication when child was feeling better was fair [$\kappa(35) = .36$, 95% CI = $-.21$, .92]. The kappa statistic for agreement on stopping medication when child was feeling worse was not computed as no patients reported this procedural error at the 3-month period.

Construct Validity

Better child ability scores for medication (PARQ-CA-medication) and exercise (PARQ-CA-exercise) were significantly correlated with the GAS in the expected direction. Pearson correlation coefficient between the GAS and PARQ-CA-exercise was strong [$r(26) = .62$, $p = .0008$], whereas the coefficient between the GAS and the PARQ-CA-medication was moderate [$r(32) = .38$, $p = .033$].

Kappa statistic for the PARQ-CA-medication and estimated medication adherence was 0.40 (95% CI: .10, .69), representing fair agreement. The percentage of observed *total* agreement between PARQ-CA-medication and estimated medication adherence was 75.6%. That is, 31 out of 41 children had scores on both measure either $\geq$80% or $<$80%.

Breaking down this distribution, 25 (60.9%) children had scores ≥80% and six (14.6%) had scores <80% on both measures.

A total of 19 children had available data on both PARQ-CA-exercise and estimated exercise adherence. This subgroup of children was older than those with missing data (11.3 v. 9.6 years, respectively). Pearson correlation coefficient between the two measures was relatively strong in magnitude ($r = .63$) and statistically significant ($p = .0037$). When we computed a kappa coefficient, using the <80% split, the obtained value was 0.36 [95% CI: −.09, .82], indicating fair agreement. Interestingly, the percentage of observed *total* agreement was 73.7%. Stated differently, 14 out of 19 children had comparable scores on both measures [i.e., 3 (15.8%) were classified ≥80% and 11 (57.9%) were classified <80% on both measures].

DISCUSSION

This study describes the PARQ, a new measure of adherence in JIA, and provides preliminary evaluation of its reliability and validity. It extends the current adherence literature specific to JIA in several important ways. First, the PARQ was developed after a thorough review of the adherence literature and incorporated important aspects of adherence in JIA. Its content validity was established by obtaining input from clinical scientists regarding clarity and relevance of items. As well, it was pretested in a sample of caregivers before administration. Second, it identifies who is responsible for different components of treatment. In our study, either the caregiver or the child was primarily responsible for treatment tasks. Shared responsibility was more common for the exercise than for the medication regimen.

Third, the PARQ provides health professionals with a multidimensional assessment of child ability and caregiver perceived helpfulness in relation to different treatment components. According to caregivers, children experienced greater difficulties and more negative reactions in response to following exercise as opposed to medication recommendations. In addition, they tended to follow the exercise program less frequently. These findings parallel those obtained in other studies involving children with JIA (Degotardi et al., 1999; Hayford & Ross, 1988; Rapoff et al., 1985). It may be that caregivers have a harder time encouraging their child to perform exercises when he or she is in pain (Degotardi et al., 1999). Another reason for lower child ability scores

for exercises may be attributed to the nature of the task (i.e., more time consuming and requiring more effort). Both possibilities merit further investigation. Although caregivers' perceived helpfulness of medication was not correlated with any of the child ability items in relation to medication, we did observe a significant association between perceived helpfulness of exercise and children's frequency of following such recommendations. This finding supports previous research showing that caregivers who believe treatment to be helpful are likely to encourage their children to persevere with recommendations (Wynn & Eckel, 1986; Schroder et al., 2002).

Fourth, the PARQ highlights potential errors in taking medication for JIA. It was disconcerting to observe that more than half of the caregivers in our study reported making errors in following medication recommendations. Errors in taking medication have also been noted in children with asthma (Celano, Geller, Phillips, & Ziman 1998; Leickly et al., 1998) and cystic fibrosis (Rusakow, Miller, McCarthy, Gershan, & Splaingard, 1998). Approaches to reducing errors in taking medications may involve verifying with caregivers (and children) whether they correctly understood the instructions, providing written protocols, and ensuring that they are capable of administering treatment, as prescribed.

Fifth, the PARQ alerts health professionals to potential barriers to treatment. Caregivers were mainly concerned about medication side-effects. Concerns about medication have also been noted as an important barrier to adherence among caregivers of children with asthma (Leickly et al., 1998). A review of adherence in JIA by Kroll et al. (1999) suggests that side effects are a particular concern given that some treatments (e.g., steroid medication) may change the child's appearance. Caregivers' concerns about medication use may also stem from an incomplete understanding of the treatment goals given the nature of the disease course. To diminish the saliency of this barrier, health professionals need to encourage caregivers to discuss their understanding of treatment. Emphasizing the risks and benefits of JIA treatment may also alleviate their concerns about medication side-effects. It is, however, important to note that patients in our clinic are given an initial education on the disease, an educational booklet, and a medication information pamphlet, all of which are discussed. Opportunity to further discuss this information and related concerns with a pediatric nurse is also provided. Therefore, the fact that caregivers identified medication side-effects as a barrier to their

child's treatment may speak to a different problem-the ability of caregivers to understand and seek out information.

In terms of the PARQ's reproducibility, the obtained ICCs for child ability and helpfulness items indicated stronger agreement over the two-week interval for exercise than for medication regimens. The lower consensus observed for child ability reflecting negative reactions toward medication and exercise regimens suggests that this aspect of adherence may be more susceptible to changes even over a relatively short interval. Substantial agreement was observed for locus of responsibility in relation to medication regimen, with agreement being moderate for exercise regimen. Perhaps children and caregivers are more likely to switch responsibility for exercise regimen over time. The observed kappa coefficients for medication-taking behaviors indicated that forgetting and being careless are stable behaviors.

Construct validity was supported for both the PARQ-CA-medication and PARQ-CA-exercise. Both composite scores were positively and statistically related to the GAS, suggesting that children with higher overall child ability are more likely to adhere to their treatment plan. These correlations were not low, which would have indicated poor validity; nor were they too high to undermine the additional value of the PARQ. Further evidence regarding its construct validity was supported by examining its concordance with estimated adherence derived from diary reports compared to actual prescriptions. The concordance between the PARQ-CA-medication and estimated medication adherence was indicative of fair agreement. The impressive observed agreement (75.6%) was, however, lowered by kappa (cf. .40). A significant and positive correlation was found between the PARQ-CA-exercise and estimated exercise adherence. This means that scores on the child ability items varied in a consistent manner with adherence scores derived from diary reports. Examining the degree of concordance between the two measures showed that the obtained kappa value (=0.36) was low despite high observed agreement (73.7%).

The reason for low kappa values in spite of high observed agreement lies in the calculation of the kappa statistic (Feinstein & Cicchetti, 1990). Correction is made for the number of instances where agreement is expected by chance. Agreement expected by chance is determined as the sum of the products of horizontal and marginal total values (in a 2×2 table) divided by N^2. If the expected agreement is large, this correction process will convert a high observed agreement into a low kappa value. In our sample, the expected agreement coefficients for medication and exercise were .60 and .58, respectively. Feinstein and Cicchetti explain that this occurs when a sample produces horizontal and vertical marginal totals that are highly symmetrically imbalanced by less or more than 0.50. This was indeed the situation in our sample; frequencies were substantially not uniform across levels within each measure. Hence, a low kappa must be weighed against the penalty imposed when a sample is not reasonably well balanced in its marginal totals.

Although these preliminary results regarding the PARQ are promising, a number of methodological limitations need to be acknowledged. First, only one project coordinator was responsible for recruiting eligible subjects, thereby making it impossible to approach all 106 caregivers who met study criteria. Out of the 33 eligible subjects not approached, some were missed, and in a few other cases, it would have been inappropriate to discuss the study at that particular visit (e.g., newly diagnosed patients who were coming to terms with the new diagnosis; patients who had too much to deal with at a particular visit; patients who came to the clinic without a caregiver). Second, socially demanding situations (e.g., completing diaries) often interpreted as an evaluation may be threatening to respondents. As a result, they may not return their questionnaires (Delamater, Kurtz, White, & Santiago, 1988). Our response rate of 73.8% can be considered satisfactory given that caregivers were asked to complete a series of measures, including six daily logs over a 3-month period. The 41 participants retained in the study did not differ significantly from their counterparts who withdrew ($n = 16$) or were dropped from the analysis due to missing data ($n = 4$) on important study variables. However, the sample retained comprised children whose cultural background was Canadian, whose mothers were highly educated, and who resided in financially secure, two-parent families. Replication of our analyses with a more diverse population is necessary before conclusions can be reached. Third, the small sample size precluded investigating for developmental differences (young children, school-aged children, and adolescents) across PARQ items. Finally, investigators must often contend with method variance when testing for validity. Method variance refers to variance attributed to the method of measurement (Larrabee, 2003). High correlations, for instance, can result from using scores derived from the same questionnaire or from using scores derived from similar methodology even though the stimuli

are different. In our study, we relied on caregiver self-reports to determine validity and reliability. This may have explained, in part, the higher correlation coefficients obtained between the PARQ-CA-exercise and the GAS ($r = .62$) and the PARQ-CA-exercise and estimated exercise ($r = .63$). These coefficients were not above .70, however, to undermine the additional value of the PARQ-exercise component.

At present there is no gold standard of measurement for pediatric populations. Investigators must therefore weigh the advantages and disadvantages of each measurement option while bearing in mind their resources. In this study, we derived estimates of adherence rates by comparing bi-weekly diary reports to actual prescriptions. Caregivers were instructed on how to complete the diaries and health professionals were provided with a standardized form that stipulated possible components of treatment and their characteristics (e.g., type, frequency, and duration of therapies). Notwithstanding our efforts to measure actual treatment-related behaviors, the completion of diaries may have heightened caregivers' awareness of children's behaviors (i.e., frequency, level of difficulty, negative reactions toward treatment), thereby altering child ability itself. Although one may argue that the use of microelectronic monitors and drug assays are less susceptible to bias or inaccurate reports provided by caregivers, these methods are not without limitations (Rapoff, 1999). Assays are influenced by individual differences in metabolic rates and recordings, and they may also reflect inadvertent behaviors such as the patient omitting or taking medications prior to assessment. Electronic monitors cannot guarantee that medication has been ingested or specific exercises were performed as recommended. Moreover, similar to diary methods, children's treatment-related behaviors could improve as a result of monitoring behaviors. In our future work, a cohort of children will be completing the PARQ. Their responses can then be compared to those obtained from their caregivers. Research with other chronic populations indicates that child reports of treatment-related behaviors and perception of who is responsible for treatment tasks differs from caregiver reports (Anderson et al., 1990; Leickly et al., 1998; McQuaid et al., 2001). By having children complete the PARQ, we will be able to reduce, to some extent, method variance.

The clinical implications of these findings can be summarized as follows. Disease management may be improved by understanding how children and caregivers handle treatment-related issues. Having caregivers complete the PARQ as part of their routine visits may assist health care providers in determining who is primarily responsible for different treatment tasks and in evaluating child ability in relation to therapies. When the child is responsible, the appropriateness of that responsibility may be discussed with caregivers in terms of the child's developmental stage, cognitive maturity, and knowledge of JIA treatment. As well, evaluation of child ability ratings may offer insights into how the child is coping with the disease and its treatment. Addressing errors in medication-taking behaviors may prevent subjecting the child to unnecessary medical tests and/or changes in medication doses. Finally, helping families deal with barriers may encourage them to follow recommendations.

In conclusion, the findings provide preliminary empirical support for the use of the PARQ in determining adherence issues in JIA. Psychometric analysis is a continuing process, which cannot be definite with a single study. Validity and reliability are evolving properties that are strengthened by continued testing of theoretical ideas and by repeated use of the PARQ in other settings and with other population groups. Future research will examine the independent influences of caregiver characteristics (distress and coping with child's illness) on child ability in relation to aspects of treatment, as well as how child characteristics (age, gender, quality of life) may modify their relative impact. This will provide a broader perspective from which to consider the 'trustworthiness' of responses derived from the PARQ in terms of their interpretation and theoretical relations with other variables (Messick, 1995).

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