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ASSESSING RELIABILITY AND VALIDITY OF A FUNCTIONAL PAIN MEASURE FOR JOINT HYPERMOBILITY

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Assessing Reliability and Validity of a Functional Pain Measure for Joint Hypermobility

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Abstract

The Pediatric Outcomes Data Collection Instrument (PODCI) is a self-reported outcome measure designed to assess upper extremity function and pain in adolescents. The purpose of this study was to determine the test-retest reliability and concurrent validity of the upper extremity function and pain portions of the PODCI (PODCI UEF and PODCI P) as an outcome measure in adolescents diagnosed with joint hypermobility (JH). The study also sought to establish the responsiveness of the PODCI subscales by calculating the standard error of measurement (SEM) and the minimal detectable change (MDC). One hundred and fifty adolescents (ages 11-18 years) with JH were recruited for the study. Test-retest reliability of both portions of the PODCI was examined with 83 participants using Lin's concordance correlation coefficient (ρ_c) There was a very high test-retest reliability for the PODCI UEF, $\rho_c = .81$ ($p < .001$) and moderate test-retest reliability for the PODCI P, $\rho_c = .68$ ($p < .001$). To establish concurrent validity, the PODCI UEF was compared to the Patient Reported Outcomes Measurement Information System Pediatric Upper Extremity-Short Form (PROMIS PUE-SF) and the PODCI P was compared to Numeric Rating Scale (NRS) using Spearman rho correlations. There was a very high correlation between the PODCI UEF and the PROMIS PUE-SF, $r_s = .80$ ($p < .001$) and a moderate inverse relationship $r_s = -.73$ ($p < .001$) between PODCI P and NRS scores. These results, indicate concurrent validity between instruments. Occupational and physical therapists may consider using the PODCI clinically and in future research.

Keywords: PODCI, joint hypermobility, pain, upper extremity function, PROMIS, NRS

Assessing Reliability and Validity of a Functional Pain Measure for Joint Hypermobility

Joint hypermobility (JH) is a chronic condition that is defined by excessive movement of the joints (Tinkle et al., 2009). Children and adolescents with JH often live with joint pain, fatigue, and poor tolerance to physical activity (Birt, Pfeil, MacGregor, Armon, & Poland, 2013). They are more likely to experience pain than their non-hypermobile peers, which can significantly impact their quality of life and participation in daily activities (Tobias, Deere, Palmer, Clark, & Clinch, 2013). Intervention provided by occupational and physical therapists is often recommended as part of a multidisciplinary approach to aid in the management of pain and improve functional limitations in this chronic condition (Cincinnati Children's Hospital Medical Center [CCHMC] Joint Hypermobility Team, 2014). However, the reliability and validity has not been established for outcome measures used to measure pain and functional performance for children with JH, limiting the ability of researchers and clinicians to confidently interpret outcomes for this population. The use of valid and reliable outcome measures may assist in planning care and identifying best practice for the management of JH.

Current literature recognizes the need for outcome measures that assess pain, function, and quality of life in adolescents within the therapy context to improve future research and clinical management of JH (Kemp et al., 2010). The Pediatric Outcomes Data Collection Instrument (PODCI) is a patient-reported outcome measure designed by the American Academy of Orthopedic Surgeons Outcomes Studies committee that is intended for use in the clinical setting (Daltroy et al., 1998). The upper extremity function subscale (PODCI UEF) measures upper extremity function and the pain/comfort subscale (PODCI P) measure the impact of pain on activity participation. The PODCI UEF and PODCI P are currently used in clinics to evaluate functional performance and pain in children diagnosed with JH; however, the validity and

reliability of the subscales of the PODCI in this population has not been established. The purpose of this study is to determine the validity and reliability of the PODCI UEF and PODCI P subscales in measuring outcomes in adolescents diagnosed with JH. Specifically, this study aimed to address the following objectives:

1. To establish the stability of the PODCI UEF and PODCI P through the use of test-retest reliability.
2. To determine the concurrent validity of the PODCI UEF by comparing its results to the Patient Reported Outcomes Measurement Information Systems, Pediatric Upper Extremity Short Form (PROMIS PUE-SF).
3. To determine the concurrent validity of the PODCI P by correlating its results to the pain scores from the numeric rating scale (NRS).
4. To establish the responsiveness of the PODCI subscales, the minimal detectable change (MDC) and the standard error of measurement (SEM) will be calculated.

Literature Review

Background

Joint hypermobility is a hereditary condition affecting the connective tissues of the body, multiple joints, and it is often characterized by pain (Schubert-Hjalmarsson, Ohman, Kyllerman, & Beckung, 2012). It is typically diagnosed using the Beighton scale criteria, in which excessive motion must be present in five or more of nine designated joints: elbows, finger metacarpals, thumbs, knees, and spine (Junge, Jespersen, Wedderkopp, & Juul-Kristensen, 2013). Joint hypermobility is a connective tissue disorder and the impact can be multidimensional, including increased pain and fatigue, muscle weakness, decreased proprioception, diminished activity tolerance, and anxiety. There is an increased incidence of depression, anxiety, and panic

disorders among people with JH (Albayrak, Yilmaz, Akkurt, Salli, & Karaca, 2015). Pain is a common complaint among individuals with JH, influencing activity and disability (Bathen, Hångmann, Hoff, Andersen, & Rand-Hendriksen, 2013). The combination of physical and psychological factors is highly correlated with disability, which is significantly present in individuals with JH (Scheper et al., 2016). Joint hypermobility is present in genetic conditions such as Marfan syndrome and osteogenesis imperfecta; however, Ehlers-Danlos, type III (EDS-III, or benign hypermobile-type) is the most common genetic condition with JH as a primary characteristic. Throughout the literature the term EDS is often used interchangeably with JH and is now considered to be the same diagnosis among experts. Uniting the diagnoses of JH and EDS-III allows for clinicians and researchers to better serve the population (Tinkle et al., 2009).

Impact of Joint Hypermobility

It is becoming more recognized in the literature that there is a correlation between JH and musculoskeletal pain (Scheper et al., 2016). One theory is that the ligament laxity causes stress and strain to the joint, which can result in acute injury of ligaments and soft tissue (Albayrak et al., 2015). Albayrak et al. (2015) completed a cross-sectional study comparing pain, depression level, fatigue, sleep, and quality of life in 115 participants with JH with 114 age-matched and gender-matched healthy controls. The group with JH demonstrated higher scores than the healthy controls on the Beck Depression Inventory (14.57, 8.37, $p < .001$) indicting more depression, increased fatigue as measured on the Checklist Individual Strength (83.97, 71.02, $p < .001$) and impaired sleep quality per the Pittsburgh Sleep Quality Index (7.53, 6.14, $p < .001$). Regarding quality of life, the group with JH reported lower physical function (66.73, 77.01, $p < .001$), lower emotional role (41.15, 72.51, $p < .001$) and higher bodily pain (52.69, 25.52, $p < .001$) on the Short Form Health Survey (SF-36) (Albayrak et al., 2015).

People with JH often demonstrate poor quality of life when compared to the general population (Bovet, Carlson, & Taylor, 2016). A cross-sectional study examined the quality of life, pain, and shoulder function among 110 participants with JH and 140 age-matched and gender-matched controls. The authors used the numeric rating scale (NRS) to assess pain, the Western Ontario Shoulder Instability Index to assess shoulder function, and Health Related Quality of Life (HRQoL) short-form to assess quality of life (Johannessen, Reiten, Løvaas, Maeland, & Juul-Kristensen, 2016). The group with JH reported higher mean pain intensity (6.4, 2.7, $p < .001$), lower mean shoulder function (49.9, 83.3, $p < .001$), and decreased mean HRQoL on the physical component scale (28.1, 49.9, $p < .001$) than the control group (Johannessen et al., 2016). Furthermore, there was higher incidences of pain reported in the shoulder in the JH group than in the control group, which the authors concluded may be a consequence of shoulder instability. The authors reported a correlation between pain, lower shoulder function, decreased physical function, as well as decreased leisure and work-related activity participation, when compared with the control group (Johannessen et al., 2016).

Adults diagnosed with JH demonstrated higher incidences of disability in mobility and activities of daily living than in the general population (Engelbert et al., 2017). Chronic and recurrent pain is a common complaint among people with JH and can lead to disability (Castori et al., 2012). Adults with JH reported significantly higher pain, decreased physical function, and increased limitations due to physical problems than same age-matched peers, which often led to disability (Rombaut, Malfait, Cools, De Paepe, & Calders, 2010). A study by Rombaut et al. (2012), compared muscle strength and endurance using isokinetic dynamometry in 43 females with JH and 43 age-matched healthy control females. The JH group demonstrated significant muscle weakness, reduced static muscle endurance, and decreased functional performance when

compared to the control group as measured by the Arthritis Impact Measurement Scales (Rombaut et al., 2012).

Joint hypermobility has been identified in up to 34% of school-aged children (Birt et al., 2013). A prospective cohort study by Tobias et al. (2013) found that children diagnosed with JH have an approximately two-fold increased risk of having musculoskeletal pain later in adolescence, specifically in the shoulder, knee, and ankle/foot. Schubert-Hjalmarsson et al. (2012) found that 20 children with JH (ages 8-16 years) reported significantly more pain in a pain diary over a two week period when compared to a control group of non-hypermobile peers ($p < .001$). Additionally, the JH group demonstrated significantly less balance ($p < .001$) when measured by the Bruininks-Oseretsky test of motor proficiency and reported lower activity levels on the frequency of participation questionnaire ($p < .001$). The children in the JH group reported a significantly higher number of days in which activity participation was limited due to pain ($p < .001$). The authors concluded that pain appeared to influence activity and participation in children with JH (Schubert-Hjalmarsson et al., 2012). Children with JH may begin to avoid physical activities that they perceive may be painful, which consequently can lead to deconditioning (Rabin, Brown, & Alexander, 2017).

Adult Intervention

Physical and occupational therapy are recommended interventions for adults and children with JH; however, managing treatment for individuals with JH is challenging for therapists due to the complexity of their symptoms (Engelbert et al., 2017). There is a paucity in research regarding intervention for adults with JH. A systematic review by Corrado and Ciardi (2018) found no randomized control trials to support rehabilitation for individuals with JH. Most studies reported were primarily investigative in nature making it difficult to specify guidelines for

therapeutic interventions (Corrado, & Ciardi, 2018). Additional research would be beneficial to guide therapy practice for individuals with JH. Focus groups from a study by Bovet, Carlson, and Taylor (2016) revealed themes of past negative therapy experiences including perceived injury and unmet therapy needs. Participants identified that caregivers frequently demonstrated lack of knowledge and that exercises were not appropriately tailored to their condition of hypermobility. Conversely, a positive therapy experience included therapists who were knowledgeable regarding JH, patient centered care, and multidisciplinary coordination across the healthcare system (Bovet et al., 2016). One pilot study found that a multidisciplinary rehabilitation program with physical and cognitive behavioral therapy was a possible intervention for women with JH (Bathen et al., 2013). Following a multidisciplinary inpatient rehabilitation with subsequent home program performance, a cohort of 12 women with JH found clinically significant improvement (2.95, 4.08, $p < .008$) in performance of activities of daily living as measured by the Canadian Occupational Performance Measure (Bathen et al., 2013).

A systematic review by Smith et al. (2013) concluded that adults with JH demonstrate reduced joint proprioception compared to the general population, which may affect coordination and activity participation. A recent study by Scheper et al. (2017) found that proprioception is an influence on the relationship between activity limitations and muscle strength. The same study also found a correlation between low muscle strength and activity limitations in EDS-HT patients. These findings support evidence for the core assumption that treatment based on muscle strengthening and increasing proprioception acuity might be effective (Scheper et al., 2017). However, higher level evidence is needed to support which therapeutic interventions are most effective (Corrado & Ciardi, 2018)

Pediatric Intervention

Treatment to address pain and functional limitations in childhood may help to limit disability in adulthood (CCHMC Joint Hypermobility Team, 2014; Engelbert et al., 2017). However, there has been limited research that has been completed with children and adolescents with JH regarding therapy intervention and management of the chronic condition. Kemp et al. (2010) completed a randomized controlled trial comparing a targeted physiotherapy program with a generalized strengthening and fitness program in 57 children (ages 7-16 years) with JH. The targeted program focused on improving joint positioning, motor control, and stability while the fitness program focused on exercises aimed at strengthening. Pain was assessed using the Visual Analog Scale (VAS) for pain as perceived by the child and the parental-VAS for the child's pain as perceived by parents. Both groups demonstrated statistical improvement in child and parent pain scores following intervention ($p < .05$); however, there was not a statistical difference between the two groups (Kemp et al., 2010).

A qualitative study found that children with JH and their parents saw improvement after completing a multidisciplinary treatment intervention that included physiotherapy. Despite recognizing the benefits of completing their prescribed home exercise program, children and parents reported difficulty with adhering to their exercises (Birt et al., 2013). Client-centered and family-centered care can help to improve outcomes and support adherence to home programs (Birt et al., 2013; CCHMC Joint Hypermobility Team, 2014). There is limited research supporting specific therapeutic interventions and protocols, despite therapy being a recommended intervention (Smith et al., 2013). There is a need for quality research to investigate the effects of therapeutic intervention including joint stabilization, strengthening, participation in physical activity, and pain management in adolescents with JH (Scheper et al., 2013)

Outcome Measures

Pain. There is not a consistent outcome measure reported in the literature that is used to assess pain in adolescents with JH. The current literature reflects the use of separate outcome measures to assess pain, function, quality of life, and strength. Pain is a primary concern for individuals with JH (Bathen et al, 2013). The Focus 17 Pain Questionnaire was used in a prospective cohort study by Tobias et al. (2013). The authors used logistic regression analysis to explore the relationship between JH and musculoskeletal pain in adolescents. Study participants with JH were more likely to have musculoskeletal pain in the shoulder (odds ratio [OR] = 1.68), knee (OR = 1.83), and ankle/foot (OR = 1.82) than their non-hypermobile peers (Tobias et al., 2013). However, the Focus 17 Pain Questionnaire has not been established as a reliable or validated assessment for the hypermobile population.

The faces pain scale was used in a randomized control trial in children with JH for participants younger than 11 years and the VAS was used for participants older than 11 years as the primary outcome measure for the interventions. The study used the additional outcome measures of the Childhood Health Assessment Questionnaire (CHAQ), parents' assessments of their child's pain, and the six-minute shuffle test to further assess quality of life, pain, and function. The authors recognized the need for more effective outcome measures within the therapy context to help improve research quality (Kemp et al., 2010).

The NRS was used in conjunction with the Functional Disability Inventory (FDI) to examine change in pain related functional disability and pain intensity in children undergoing cognitive behavioral therapy (Lynch-Jordan et al., 2014). It is expected that there is a correlation between pain and function. However, the authors noted a statistically significant difference in function on the FDI, but not in pain on the NRS following the treatment (Lynch-Jordan et al.,

2014). The NRS is commonly used clinically and in research to measure pain intensity in children and adolescents with chronic pain including headaches, abdominal pain, complex regional pain syndrome, and musculoskeletal pain (Castarlenas et al., 2017; Ruskin et al., 2014; Sheehy et al., 2015). However, it does not measure how that pain may influence participation in daily activities or how it may contribute to disability. Accordingly, the current study seeks to establish the PODCI P as a reliable and valid outcome measure that assesses the interference of pain on function for adolescents with JH.

Function. There is a lack of valid and reliable outcome measures to assess upper extremity function in adolescents with JH. The Pediatric Quality of Life Inventory 4.0 (PedsQL) is a pediatric and adolescent quality of life assessment that was found to have strong test-retest reliability, intraclass correlation coefficient (ICC) = .82, in a small sample of 10 children with JH. However, the sample size was not large enough to generalize the results to the larger population of children with JH (Fatoye et al., 2012). The PedsQL offers a score on global quality of life, but it does not specifically address pain and has limited questions on physical function. There is a need for patient reported outcome measures for adolescents with JH that can be completed quickly within the therapy visit and that can measure gains in function as well as reduction in pain and its interference with activity.

Pediatric Outcomes Data Collection Instrument. Patient-reported outcome measures can be useful instruments to assess baseline function, improvement in function, and to guide clinical practice (Kempert, Benore, & Heines, 2017). The PODCI is a patient-reported outcome measure designed by the American Academy of Orthopedic Surgeons Outcomes Studies committee for use in children and adolescents, aged 2-18 years with a variety of musculoskeletal conditions including cerebral palsy, brachial plexus palsy, and congenital differences (Bae,

Waters, & Zurakowski, 2008). It consists of three different forms: pediatric form (for ages 2-10 years) that is filled out by a parent/caregiver; adolescent parent report form that is filled out by parent/caregiver of adolescents ages 11-18 years; and the adolescent self-report form for ages 11-18 years. The PODCI measures outcomes across eight domains: upper extremity and physical function, transfer and basic mobility, sports/physical function, pain/comfort, treatment expectations, happiness, satisfaction with symptoms, and global functioning. Each scale or domain yields a standardized score between 0-100. Test-retest reliability, sensitivity to change, and construct validity were established during development (Daltroy, Liang, Fossel, & Goldberg, 1998). Testing was completed in multiple institutions among a variety of musculoskeletal conditions including scoliosis, dysplasia, cerebral palsy, juvenile arthritis, leg-length discrepancies, developmental delays, and abnormal gait (Daltroy, et al., 1998). In the adolescent self-report questionnaire, the test-retest reliability was found to be high (ICC = .75 to .92). Construct reliability was established by comparing the PODCI to the CHAQ. A strong positive correlation was found indicating that the PODCI produces similar results when measuring comparable constructs (Daltroy et al., 1998).

Hunsaker et al. (2002) found the PODCI demonstrated high internal consistency, as well as discriminant validity in the general population (ages 2-18 years). Multi-trait scaling techniques were used to examine reliability and item discriminant validity across all 11 scales (Hunsaker et al., 2002). The PODCI demonstrated high internal consistency (Cronbach's alpha exceeded .80). In multi-trait scaling, discriminant validity examines correlations of items of the scales of the assessment with items of other scales. Discriminant validity is reached when there is ≥ 80% of the item to scale correlations in the total data set and within each individual scale (Hunsaker et al., 2002). Cronbach's alpha range was .94 - .98 ($p < .05$) for the upper extremity

portion of the PODCI, indicating that item discriminant validity was achieved (Hunsaker et al., 2002). The mean scores from the study were high across scales in the normative sample, however also demonstrated meaningful variability. Therefore, the authors concluded that the PODCI assessments will be utilized more effectively in populations with poor health. In an analysis among children with congenital limb differences, aged 2-18 years, the mean score for upper extremity function was significantly lower than in the normal population (76.6, $p < .05$; 98.7, respectively) (Wall, Shen, Roberts, & Goldfarb, 2016; Hunsaker et al., 2002).

The utility and reliability of the PODCI was assessed in children between 2 and 18 years of age with acute hand and wrist injuries. The authors found that it demonstrated good internal consistency. The Cronbach's alpha for the PODCI UEF was .89 and .87. The study established that the PODCI UEF and PODCI P were responsive to change in function, with a standardized response mean of 0.42 (effect size 0.55) and 1.27 (effect size 1.37), respectively. The PODCI was also found to discriminate upper extremity function between children with acute hand and wrist injuries from children in the general population and between children with different injury locations. Furthermore, administering the PODCI did not impact treatment time in the clinic or patient satisfaction; therefore, it may be a useful outcome measure clinically and for research purposes (Kunkel, Eismann, & Cornwall, 2011).

The clinical utility of the PODCI as an outcome measure, as well the established reliability and validity in multiple populations, make it a good option for the present study. In a study in adolescents with upper extremity conditions, the PODCI was found to demonstrate high correlation ($r = .85$) with the Pediatric QoL Inventory, indicating construct validity (Mahan et al., 2014). Similarly, a study by Pencharz, Young, Owen, and Wright (2001) reported good construct validity of PODCI by correlating it with the Child Health Questionnaire Parent Form and the

Activities Scales for Kids (ASK). The study found that the PODCI was effective in measuring physical function and pain in children with disability, however the full PODCI questionnaire (108 questions) was not always feasible in the clinic setting.

A recent retrospective study explored the relationships between functional disability and depression symptoms in children and adolescents with chronic pain. Bivariate correlations were separately performed between the subscales of the PODCI (global functioning, upper extremity and physical functioning, transfer and basic mobility, and sports and physical functioning) and the Children's Depression Inventory (Broadbent, Bertino, Brooke, Fuller-Tyszkiewicz, & Chalkiadis, 2017). The authors found that depression and functional disability were not correlated among any of the four subscales, suggesting that the PODCI may be a useful measure of functional disability without the confounder of symptoms of depression in pediatric patients with pain (Broadbent et al., 2017).

The PODCI has been established as a feasible, reliable, and valid clinical assessment in several orthopedic conditions including cerebral palsy, brachial plexus palsy, upper extremity acute conditions, and musculoskeletal conditions (Bae, Waters, & Zurakowski, 2008; Kunkel, Eismann, & Cornwall, 2011; Mahan et al., 2014; Pencharz et al., 2001). However, the utility of the PODCI has not been examined in adolescents with JH. Therefore, it is hypothesized that the adolescent self-report PODCI, specifically the upper extremity and pain/comfort scales, may be a reliable and valid outcome measure to use with adolescents with JH. The PODCI UEF and PODCI P are currently being used as an outcome measure by therapists at CCHMC. The PODCI UEF measures how easy or hard it is to complete daily tasks such as writing, combing hair, and opening containers. The PODCI P measures the impact of pain on function over the last week, as well as how pain may have interfered with participation in activities. The entire PODCI consists

of a large number of items and may take away from treatment time in order to complete it (Mahan et al., 2014; Quatman-Yates et al., 2013). Accordingly, the aim of this study is to establish the PODCI UEF and PODCI P as reliable and valid outcome measurements in adolescents with joint hypermobility. Doing so may help to guide future research and clinical practice in the management of joint hypermobility.

Method

Study Design

An observational study using a cross-section design was conducted to establish validity and reliability of the PODCI. The study took place from June 1, 2018 to January 31, 2019 at CCHMC. Prior to participant recruitment, the study was approved by the Institutional Review Board (IRB) at CCHMC and a reliance agreement was executed with the Human Research Protections Program at the University of Indianapolis.

Participants

A convenience sample of adolescents referred by a physician or nurse practitioner for occupational therapy and/or physical therapy at CCHMC to address pain or difficulties related to their JH were recruited for the study. The treating therapists and the primary researcher (C. S.) identified potential participants prior to their therapy visit through chart reviews completed as standard of care. To participate in the study, individuals must have met the following inclusion criteria:

- Age 11-18 years at the time of recruitment
- Diagnosed with joint hypermobility or Ehlers-Danlos, type III
- Referred by a physician to occupational and/or physical therapy due to functional limitations and/or pain that are associated with their JH

- Speak English as their first language
- Ability to read and comprehend English at the 5th grade level
- Must have a parent or legal guardian present during the treatment session if the adolescent is 17 years of age and under

Individuals were excluded from the study if they had a formal diagnosis in their medical chart of another musculoskeletal disorder, intellectual disability, pregnancy, a co-existing disabling condition or joint diseases, including but not limited to arthritis or lupus.

Sample size. A sample size calculation was conducted using the formula provided by Walter, Eliasziw, and Donner (1998) for reliability studies using the ICC. It is estimated that a minimum of 117 participants are required to established the test-retest reliability of the PODCI, with an alpha of .05, power of .80, acceptable reliability of .70 and expected reliability of .90 (Portney & Watkins, 2000). To account for possible attrition, the sample size was increased by 20% for a final sample size of 147 participants.

Data

Quantitative data were collected through Qualtrics (https://www.qualtrics.com/), a secure, online survey program. The initial PODCI, PROMIS, and NRS scores were collected during the participants' therapy visits. The second PODCI assessments were completed between 5-14 days following the initial visit via a link that was sent to the participant through text message or email. In addition, participant age and gender were also collected at Time 2. Data from both time periods were downloaded from Qualtrics.com into a Microsoft Excel file by the primary researcher and collated using the unique ID entered by the participant. Once the scores for each of the instruments were calculated, the data were exported into a statistical software program for analysis.

Instruments

The Pediatric Outcomes Data Collection Instrument. The PODCI is a self-reported outcome measure designed by the American Academy of Orthopedic Surgeons Outcomes Studies committee that was intended for use in the clinical setting (Hunsaker et al., 2002). The PODCI measures outcomes across eight domains: upper extremity and physical function, transfer and basic mobility, sports/physical function, pain/comfort, treatment expectations, happiness, satisfaction with symptoms, and global functioning. Each scale or domain yields a standardized score between 0-100. In a population of typically developing children aged 2-18 years, the PODCI demonstrated high internal consistency as reported above and item discriminant validity when compared with the Disabilities of the Arm, Shoulder, and Hand (DASH) assessment (Hunsaker et al., 2002). In children and adolescents with acute wrist and hand conditions, the full version of the PODCI demonstrated internal consistency (Cronbach's alpha = .89) and responsiveness to change (Kunkel, Eismann, & Cornwall, 2011).

This study used two subscales of the PODCI, the upper extremity function (UEF) and pain/comfort (P). This modified version of the PODCI consists of 11 questions designed to be used with children ages 11-18 years. Eight questions assess upper extremity function. The participant is asked to assess how easy or hard the following tasks are: writing with a pencil, pouring a half gallon jug, lifting heavy books, fastening buttons, using a fork or spoon, opening a jar that has already been opened, combing hair, and turning a doorknob. The activities are rated on a four-point Likert-like scale: 1 (easy), 2 (a little hard), 3 (very hard), and 4 (can't do at all). A score of 100 indicates no functional difficulties, while a score of 0 indicates that the user is unable to complete the tasks. The mean (with standard deviation in parentheses) in pediatric normative sample is 98.71 (4.93) (Hunsaker et al., 2002). A retrospective study by Gates and

Campbell (2015) compared the norms established by Hunsaker, et al. (2002) to children and adolescents to evaluate the impact of a variety of comorbid conditions, age, and gender on the PODCI subscales using independent samples *t* tests. In that study, the mean for PODCI UEF in individuals with a comorbidity that was currently limiting was 93.1 (14.8). There was not a significant difference between the scores, $F(1, 5047) = 0.003$, $p < .896$. The comorbid conditions included arthritis, eating disorders, asthma, learning difficulties, heart conditions, vision and hearing impairment (Gates & Campbell, 2015).

Three questions address how much pain interfered with the individual's activity over the last week. A score of 100 indicates no pain, while a score of 0 indicates that pain severely interfered with participation in activities. The mean in pediatric normative sample is 89.31 (14.79) (Hunsaker et al., 2002). In children and adolescents with a currently limiting comorbidity, the mean for the PODCI P was 77.0 (22.6). There was a significant difference between the normative sample and the sample with comorbidities, $F(1, 5047) = 29.75$, $p < .001$) (Gates & Campbell, 2015).

The PODCI is calculated as follows:

- Upper extremity function score = [(4 - x)100]/3;

  x = mean of questions 1, 2, 3, 4, 5, 6, 7, 8
- Pain score= [(4 - (x - 1))100]/4; x = the mean of y, z, and question #11; y = [(4 - the response to question #9)4]/3 +1; z = [(the response to question #10 -1)4]/5+1

Patient Reported Outcomes Measurement Information System, Pediatric Upper Extremity Short Form v2.0. The PROMIS is set of person-centered measures, initiated and supported by the National Institute of Health to aid in the evaluation of physical, mental, and social health across ages and health conditions (Irwin et al, 2012). The pediatric set is self- report

for ages 8-17 years and measures across five health domains of physical function, pain, fatigue, emotional health, and social health. The five domains have been further defined into multiple forms for assessments to be used in the clinical and research environments. The PROMIS PUE-SF has demonstrated test-retest reliability among 54 participants with a correlation of .71 and good internal consistency with an alpha of .63 (Varni et al., 2014). It demonstrated good construct validity with the PODCI ($r = .70$, $p < .001$) and the DASH ($r = .80$, $p < .001$) in children with congenital hand differences (Waljee, Carlozzi, Franzblau, Zhong, & Chung, 2015). The PROMIS PUE-SF consists of eight questions to assess daily upper extremity function. Participants are asked to rate how much trouble it is to do the following items: button a shirt or pants, open a jar, open the rings in a school binder, pour a drink from a full pitcher, pull a shirt on over my head by myself, pull open a heavy door, put on shoes, use a key to unlock a door. The items are rated on a five point Likert scale 1 (not able to do), 2 (with a lot of trouble), 3 (with some trouble), 4 (with a little trouble), and 5 (with no trouble). There are two items that have collapsed scoring responses (button shirt or pants and pull shirt overhead). The PROMIS UEP and PROMIS P are scored using T–scores, mean of 50 and a standard deviation of 10; therefore, a score of 40 would indicate upper extremity function 1 standard deviation below the mean while a score of 60 would indicate upper extremity function 1 standard deviation above the mean (Thissen, et al., 2015). It is recommended to use the Health Measures Scoring Service to score the assessment, found at https://www.assessmentcenter.net/ac_scoringservice. However, it can also be calculated by hand, converting raw scores into standard scores using scoring tables in the PROMIS Physical Function Scoring Manual found at http://www.healthmeasures.net/administrator/components/com_instruments/uploads/PROMIS%20Physical%20Function%20Scoring%20Manual.pdf.

The Numerical Rating Scale. The NRS is commonly used by many disciplines across ages and diagnoses to measure an individual's pain (Brevik et al., 2008). A systematic review completed by Castarlenas, Jensen, von Baeyer, and Miró (2017) found the NRS to be a reliable and valid pain assessment tool for children over 8 years of age. However, it should be noted that only one of the sixteen articles reviewed established test-retest reliability in the measure. It was established using the Bland-Altman method and showed a high agreement, limits of agreement of 95% CI [0.90, 1.20] (Castarlenas et al., 2017). Convergent validity was established in children, ages 8-17 years, with chronic pain by comparing it to the Coloured Analog Scale (CAS). The authors found that the NRS met criteria for convergent face validity ($r > .30$ to .50) and correlated with the CAS on all four pain levels ($r = .58$ to .68, all $p < .001$). Discriminant validity was examined by comparing the magnitude of the correlations for NRS and CAS usual pain ratings with the correlation between NRS and Facial Affective Scale (FAS) using Fisher's Z-transformation test (Ruskin et al., 2014). The magnitude of the correlation between NRS and FAS was found to be significantly lower than that between the NRS and CAS ($Z = 2.84$; $p = .005$) (Ruskin et al., 2014).

Procedures

Recruitment. Potential participants were identified after physician or nurse practitioner referral to occupational therapy and/or physical therapy. Treating therapists completed chart reviews to assess for inclusion and exclusion criteria at the time of their regularly scheduled therapy appointment. Potential participants must have met the inclusion criteria at that time to be invited to participate in the study. The treating therapist or the primary researcher verbally reviewed the consent document with the potential participants and their parent/legal guardian. They were also given a written description of the study to review. They were given time to ask

questions and make a decision about being in the study. Formal consent was waived because data were de identified and the study posed minimal harm. Participants were considered to be in the study if they agreed to provide contact information for the study.

Data collection. Participants who agreed to be in the study completed the following:

- Wrote down an email or phone number so that the survey could be sent to them through text or email. The parent/guardian was also able provide their own email address or phone number so that they could be copied on the email or text if they chose. Writing down their email and/or text and giving it to the treating therapist implied consent to participate in the study.
- The participant used a unique six character letter/number code when filling out the PODCI that was used to match their first and second assessments. The code consisted of the first two letters of their middle name, followed by the first two letters of their mother's first name and the first two numbers of their home address. Some children chose a different six character code when completing the PODCI. This code was used on all data collection documents in Qualtrics.com and was not linked to the participants name or contact information.
- Participants filled out the PODCI (both subscales), PROMIS, and NRS through Qualtrics.com at their initial therapy visit.
- A link to the online PODCI was emailed or texted to participants 5 days following their initial therapy visit. Their parent/legal guardian was copied on the email/text when requested; however, participants and their caregiver were instructed that only the adolescent should complete the assessments. A description of the study

was included in the email or text and a copy of the informed consent form was included as the first page of the Qualtrics questionnaire.

- A reminder email or text was sent 10 days following their initial therapy visit. Participants were instructed to disregard the message if they had already completed the questionnaire through Qualtrics.com. If it was completed an additional time, the additional assessment was deleted and not collected as part of the data.
- Participants completed the PODCI within 14 days of their initial therapy visit through the secure website (Qualtrics.com) using their unique code to maintain their anonymity. Assessments completed after 14 days of the initial visit were deleted and not used in the study.
- The completed assessments were paired with each other using the six digit code provided by the participant and given a unique study number. Data were downloaded from Qualtrics.com. Only those approved to work on the research study had access to the data.

Data Analysis

Descriptive statistics were used to describe the baseline characteristics of participants. None of the Time 1 outcomes data were normally distributed; therefore, non-parametric tests were performed. In examining test-retest reliability, agreement between the PODCI UEF at two time points and the PODCI P assessments at two time points were assessed using Lin's concordance correlation coefficient ([Lin's C] Lin, 2000). A statistical calculator was used to calculate Lin's C (www.niwa.co.nz/node/104318/concordance). Cronbach's alpha was calculated to assess for internal consistency. A Cronbach's alpha level 0 is the lowest score, while a score

of 1 is the highest. An alpha of .70 is the minimal acceptable level for internal consistency for health professions research (Portney & Watkins, 2014). Concurrent validity for the PODCI upper extremity function subscale was explored by using the Spearman rho correlation test to determine if there was an association between the PODCI UEF and the PROMIS PUE-SF. The raw scores from the PROMIS PUE-SF were used rather than the normative scores as the PODCI UEF does not have normalized scores. The Spearman rho test was also used to determine if there was an association between the PODCI P subscale and the NRS.

The following correlation scale for health professions was used for Lin's C and Spearman rho: A score of 1.00 indicates perfect correlation, .80 - .99 = very high, .60 - .79 = moderate, .30 - .59 = fair, .29 - .10 = poor, while a score of 0 indicates no correlation (Altman, 1991). The study sought to establish test-retest reliability of at least .90 and validity of at least .70, consistent with instrumental reliability in health professions (Portney & Watkins, 2014). The minimal detectable change (MDC) was calculated using the standard error of measurement (SEM) ($1.96*SEM*\sqrt{2}$). The SEM [$SD * (\sqrt{(1 - p_c)}$] was calculated using Lin's C; however, it should be noted that the data were not normally distributed which may impact the results. The Shapiro-Wilk test was used to determine whether data were normally distributed. All data were analyzed using IBM SPSS Statistics for Windows, Version 25.0 (IBM Corp., Armonk, NY). All tests were two-tailed, and an alpha level of less than .05 was considered to be statistically significant.

Results

A total of 150 participants were recruited during regularly scheduled outpatient occupational and physical therapy visits from a pediatric hospital in the Midwest. The participants ranged in age from 11 through 18 years, with a median age of 15 years. The study recruited male and female participants, with 80% being female. This is consistent with JH being

more prevalent in females (Smith et al., 2013). Of the 150 participants, 93 completed the PODCI a second time within the 14 day time frame. Ten PODCI surveys were excluded due to not having an ID code that matched the ID code of an initial assessment. Therefore, data on the 83 (55.3%) participants are reported for the test-retest results. For validity testing, all 150 participants completed the PODCI UEF, PODCI P, PROMIS PUE-SF, and NRS.

Reliability. For the PODCI UEF portion, the Lin's C correlation indicated very high relative test-retest reliability (p_c = .80). Absolute reliability was also established with no statistically significant difference found between Time 1 and Time 2 scores (Z = -1.15 p = .25). Cronbach's alpha was calculated using the Time 1 and Time 2 scores. The internal consistency among the 8 questions was assessed to be high with a Cronbach's alpha value of .81 for Time 1 and .76 for Time 2. The Lin's C for the PODCI P indicated only moderate relative test-retest reliability (p_c = .68). However, there was not a statistically significant difference between Time 1 and Time 2 scores (Z = -1.78, p = .075), establishing absolute test-retest reliability of the subscale. Internal consistency among the three questions was assessed to be high with a Cronbach's alpha value of .88 for Time 1 and .83 for Time 2. Refer to Table 1 for the descriptive statistics for all Time 1 and Time 2 results for both time periods.

Validity. The Spearman rho correlation between the PODCI UEF and the PROMIS PUE-SF was .79 (p < .001), indicating moderately high correlation between the two measures (Figure 1). There was also a moderate, negative correlation between the PODCI P assessment and the NRS, r_s = -.73; p < .001 (Figure 2).A lower score on the PODCI P indicates greater pain, while a lower score on the NRS indicates less pain; therefore an inverse relationship was expected.

Minimal detectable change. The study also sought to establish the responsiveness of the PODCI by calculating the minimal detectable change (MDC) using the SEM. The MDC is the

smallest change that can be considered above the measurement error at a given level of confidence (Copay, Subach, Glassman, Polly, & Schuler, 2007). The confidence level was set at 95% and Time 1 data were used. For the PODCI UEF, the SEM was 6.99 with a MDC of 19.38. For the PODCI P, the SEM was 14.37 and the MDC was 39.83. This indicates that a change above these values would be 95% likely to be a true change verses a change that is caused by an error.

Discussion

This study sought to establish test-retest reliability of the UEF and pain interference portions of the PODCI in adolescents with JH. There is a lack of reliable assessments that measure upper extremity function and the impact of pain in this population. This study established an a priori correlation coefficient of .90, however this was not achieved. During its development, test-retest reliability was established for the PODCI UEF subscale (r = .96) and the PODCI P subscale (r = .89) in 30 adolescents with various chronic conditions. The test-retest assessments were taken 1-2 days apart (Daltroy et al., 1998). The smaller sample size, various diagnoses that did not include JH, and the differences in time frame may have impacted differences in the test-retest reliability between the original study and the current one. Using a convenience sample of participants currently undergoing treatment for function and pain may have also contributed to the non-normal distribution of the data which may have also biased the results. Some adolescents with JH may experience upper extremity dysfunction and pain, while others may not. This may depend largely on which joints are hypermobile, as well as the individual's unique daily activities. Some of the adolescents in the sample were undergoing occupational therapy to address upper extremity dysfunction and pain. However, some of the adolescents were not reporting upper extremity pain or dysfunction and therefore, were only

receiving physical therapy and not occupational therapy. This difference in participants may have also contributed to the non-normal distribution of data.

The PODCI UEF demonstrated better test-retest reliability than the PODCI P. It is worth noting that pain in a chronic condition such as JH can vary day-to-day which may affect the test-retest reliability in an outcome measure. The participants took the PODCI retest between 5-14 days following therapeutic intervention. It is possible that the individuals experienced changes in pain related to their condition or to their therapy visit during this time frame. The PODCI P accounts for the impact of pain on participation in activities over the last week versus the NRS that measures only the intensity of pain. Using the PODCI P along with the NRS may help clinicians to assess the impact of interventions on meaningful activities that are not part of the PODCI UEF, such as extracurricular activities and social participation. Therefore, the PODCI P may be a useful tool in measuring the impact of pain on activities over time following therapeutic intervention.

The PODCI was designed to be used as an outcome measure for chronic musculoskeletal conditions and has demonstrated test-retest reliability, validity, and responsiveness to change in other populations, making it a feasible option to use in the clinic and research settings (Kunkel, Eismann, & Cornwall, 2011). It is worth noting that reliability was established as well as a strong correlation between Time 1 and Time 2. This is clinically relevant because there is currently not an outcome measure for upper extremity function and pain that has established test-retest reliability among adolescents with JH.

Concurrent validity. This study sought to establish concurrent validity between the PODCI UEF and PROMIS PUE-SF and between the PODCI P and the NRS with an $r > .70$. This was achieved in both measures. The findings have implications that the PODCI may be a

useful tool in the clinic to assess upper extremity function and pain in adolescents with JH. There is currently not an outcome measure that has been established as valid in adolescents with JH.

Minimal detectable change. The sample did not demonstrate a normal distribution, as determined by the Shapiro-Wilk test of normality. The SEM is meaningful when calculated from a normal distribution; therefore, the SEM and MDC for this study should be interpreted with caution. The lack of normally distribute data could account for the large values. In addition, there was variation in how long participants were in treatment when they completed the PODCI. Some were just beginning therapy while others had been in therapy for several months. It is possible that those in treatment longer were experiencing less functional limitation and pain than those just beginning treatment. This could account for the non-normal distribution and large SEM and MDC. To determine responsiveness of the instrument to detect change, it would have been better to collect the scores for all participants at the onset of treatment, then again after a specified number of visits or time frame.

Not all individuals with a diagnosis of JH experience upper extremity dysfunction and/or pain. However, it is common for people with JH to have glenohumeral instability (Scheper, et al., 2013) and shoulder dysfunction (Johannessen et al., 2016). Using the PODCI UEF can help primary care providers, geneticists, and rheumatologists to identify potential areas of upper extremity dysfunction related to shoulder instability or hypermobility in the upper extremities and refer to occupational and/or physical therapy as indicated. A score of 100 on the PODCI is indicative of normal upper extremity function. Therefore a score below 90 would reveal decreased levels of function when compared to the normative sample (Haynes & Sullivan, 2001) and would suggest the need for therapeutic intervention. The PODCI UEF provides information on upper extremity function across different domains of participation, such as activities of daily

living, instrumental activities of daily living, and school which may help guide clinicians when planning treatment. Clinicians can use the Likert-like scale responses to help adolescents identify the barriers to completing the tasks, such as decreased strength or pain. Using the PODCI UEF to identify and discuss areas of dysfunction can help to improve collaboration and client centered care.

The PODCI P may help clinicians better understand how pain has impacted participation in meaningful occupations rather than the intensity of their pain at a particular moment. Children with JH report higher instances of pain that appears to limit activity and leisure participation (Schubert-Hjalmarsson et al., 2012). It is recommended clinicians treating pain to gather a comprehensive picture to help identify what is contributing to the pain (Rabin, Brown, & Alexander, 2017). The PODCI P can help clinicians gather more information about the impact of pain as well as what may be contributing to it. Additionally, the PODCI has previously been found to discriminate between the functional disability and depression that can be present in children with pain (Broadbent et al., 2017).Clinicians can use the PODCI P answers to guide questions on which areas of participation have been impacted, as well as guide subsequent treatment planning, activity modifications, and exercise program.

The NRS has been established as a reliable and valid measure of pain; however, it measures the intensity of pain at one moment in time and does not capture the impact or interference of pain on activity. To effectively help adolescents manage pain, more clinical information is needed to understand the nature of the pain and its impact (Rabin et al., 2017). The NRS and the PODCI P demonstrated only a moderate correlation which is not surprising since they are measuring different aspects of pain. Pain is complex and variable in chronic conditions and may require an assessment in addition to the NRS to better understand the impact

of pain on daily activities. Due to the inconsistent nature of chronic pain, it may be difficult to use the NRS alone as an outcome measure to assess the effectiveness of therapeutic interventions aimed at lessening pain during functional activities. The PODCI P allows clinicians to understand more about how the intensity of pain is impacting the adolescent's participation in daily activities. For example, following an intervention, the NRS pain intensity may decrease, however pain may still impact or limit participation in meaningful activities. The NRS when used alone may capture pain that is happening during their clinic visit, however may not be reflective of pain that occurs during the school day or a leisure activity. Using the PODCI P in addition to the NRS can help clinicians better understand the pain the adolescent is having which may help them develop a more client-centered intervention. The NRS is routinely used across professions in many different settings and may be beneficial to use in conjunction with the PODCI P in assessing pain and changes in how pain impacts participation daily activities over time.

The PODCI was primarily designed for children and adolescents with musculoskeletal conditions, such as JH (Quatman-Yates et al., 2013; Daltroy et al., 1998). Adolescents with scores below 80 - 90 on the PODCI indicated a difference from the normative population and therefore, may benefit from an intervention targeted at improving upper extremity function and pain. Conversely, adolescents with scores above 80 - 90 on the PODCI indicate upper extremity function and pain that is normal and may not benefit from therapeutic intervention (Haynes & Sullivan, 2001). The PODCI did appear to detect dysfunction and pain in adolescents with JH. The mean score for UEF in a normative sample was 98.71 (Hunsaker et al., 2002), while the median score for UEF in the current study was 87.50. The mean score of the PODCI P in a normative sample was 89.31. The median score in the current study was 49.44. It is important for

clinicians to use outcomes to guide practice and intervention. The first step in client-centered care is identifying an area of concern and then determining which interventions may be appropriate for each adolescent. Using a self-report measure, such as the PODCI UEF and PODCI P may be a good first step in the process.

The PODCI UEF and PODCI P instruments, when combined, are a total of 11 questions and are easy to administer in the clinic setting. Furthermore, both subscales have already been used in research with children with a variety of conditions, including brachial plexus injury, cerebral palsy, acute injuries, and limb differences. The PODCI UEF is comparable to the PROMIS PUE-SF in length, design, and content. The PROMIS PUE-SF did not demonstrate the ceiling effect that the PODCI UEF did with the same population. This could be because the PODCI was designed for adolescents with existing dysfunction and not for the general population. The PROMIS PUE-SF uses a T-score calculation with standard deviations that help to define difference from the mean, which may help it to be more sensitive to change. Many adolescents with JH report difficulty with writing and it is a question on the PODCI UEF. The PROMIS PUE-SF does not have a question regarding writing with a pencil. The PROMIS does have a pediatric pain/comfort scale short form in addition to the PUE-SF. However, it is eight questions and when combined with the eight questions from the PROMIS PUE-SF, it would take longer for an adolescent to complete. Additionally, more research is needed to determine the effectiveness of these scales as outcome measures. Neither the PROMIS PUE-SF nor the PROMIS pain interference scale has been established as valid in adolescents with JH. There is a need to establish the reliability and validity of the PROMIS scales in adolescents with JH to consider them as potential outcome measures to use clinically and in research.

It has been established that adolescents with JH often live with joint pain that impacts their physical activity and quality of life (Birt et al., 2013; Tobias et al., 2013). Therapy is often recommended as part of the treatment process and the type of intervention may be left to the treating therapist to decide. It is agreed upon in the literature that more research is needed regarding which therapeutic interventions are the most effective for adolescents with JH. A valid and reliable outcome measure is necessary for this population to help guide clinical practice and research. Using a reliable outcome measure consistently in research is one step towards improving research and clinical practice in improving pain and function in adolescents with JH. This study established that the two PODCI subscales, PODCI UEF and PODCI P, demonstrate moderate to high test-retest reliability and concurrent validity. The combined subscales are a total of 11 questions and easy to use in the clinic setting. Furthermore, it has been previously established to be responsive to change, which adds value as an outcome measure both in research and clinically (Kunkel, Eismann, & Cornwall, 2011). Therefore, the PODCI may be useful as an assessment tool as well as an outcome measure for occupational and physical therapists when treating adolescents with JH.

Limitations

The study aimed to recruit 117 participants to establish test-retest reliability. A total of 150 participants were recruited; however, only 83 completed the test-retest portion of the study. All of the participants were receiving occupational and/or physical therapy, but the length of time receiving treatment varied. Some participants may have been receiving therapy for 6 months, while others may have just been referred to therapy. Some participants were receiving therapy for reasons other than upper extremity dysfunction, or were receiving only physical therapy. This may account for 40% of the participants having an UEF score greater than 90,

while 10% of the participants had a P score greater than 90. Additionally, the PODCI was designed for children with orthopedic or musculoskeletal difficulties; therefore, there is a ceiling effect in the scores among children without orthopedic difficulties (Haynes & Sullivan, 2001). Future studies may consider excluding scores of 100, or only recruiting participants who are receiving occupational therapy. Some of the participants had been given the PODCI UEF and PODCI P prior to participating in the study as part of their regular therapy visit, which may have created a bias. The potential bias along with the variation in therapy visits may have also contributed to the non-normal distribution. Future studies may consider establishing responsiveness by administering the PODCI UEF and PODCI P prior to the onset of therapeutic intervention, then again after a controlled number of visits. Lastly, Daltroy et al. (1998) established test-retest reliability by administering Time 1 and Time 2 within 1-2 days, and it may have been better to follow a similar format in this study due to the chronic nature of the diagnosis and the variability of pain.

Conclusion

The UEF and pain interference subscales of the PODCI demonstrated moderate to high reliability and validity in adolescents with JH. Additionally, it has been previously established to be a feasible measure to use in the clinic setting (Hunsaker et al., 2002; Kunkel, Eismann, & Cornwall, 2011) and did not impact treatment time during this study. As the sample in this study did not have a normal distribution, more research is recommended to establish the responsiveness of these subscales in adolescents with JH. It is also recommended to continue to research the utility of the PODCI UEF and PODCI P as an outcome measure in adolescents with JH. While this study established concurrent validity between the PODCI UEF and the PROMIS PUE-SF, it would be helpful to determine if both are as responsive to change in function in

adolescents with JH. Likewise, it is recommended that the validity of the PODCI P in this population be compared to another pain interference instrument like the PROMIS Pain Interference scale. Despite the need for further validation of the PODCI subscales, given the lack of available valid and reliable outcome measures for this population, occupational and physical therapists may consider using the PODCI UEF and PODCI P in conjunction with the NRS clinically and in research. Primary care physicians, rheumatologists, geneticists, psychologists, and nurse practitioners may also consider using the PODCI UEF and PODCI P to determine the need for referrals for therapy. It is also recommended that clinicians continue to assess reliability and validity in outcome measures for adolescents with JH to continue to advance clinical practice and research.

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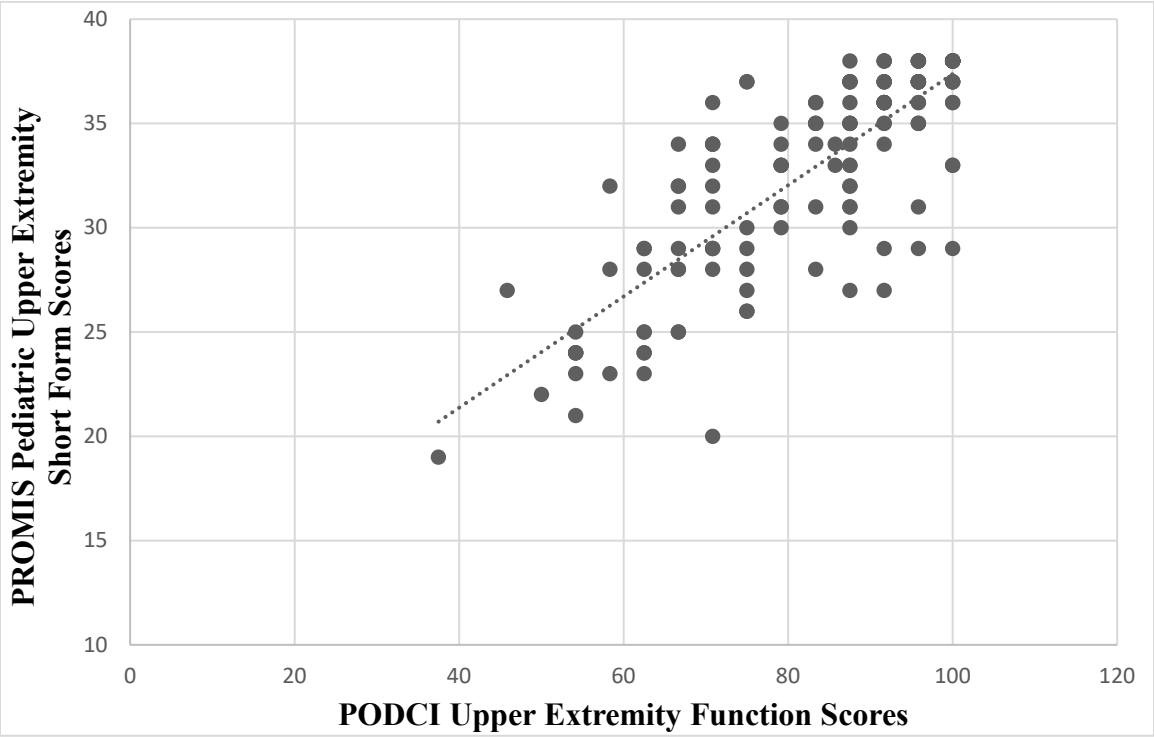
Table 1

Descriptive Statistics for Test Retest Reliability (N = 53)

PODCI	Time 1	Time 2
	Mdn (IQR)	*Mdn* (IQR)
Upper Extremity Function	87.50 (25.00)	83.33 (25.00)
Pain	49.44 (41.1)	58.89 (32.78)

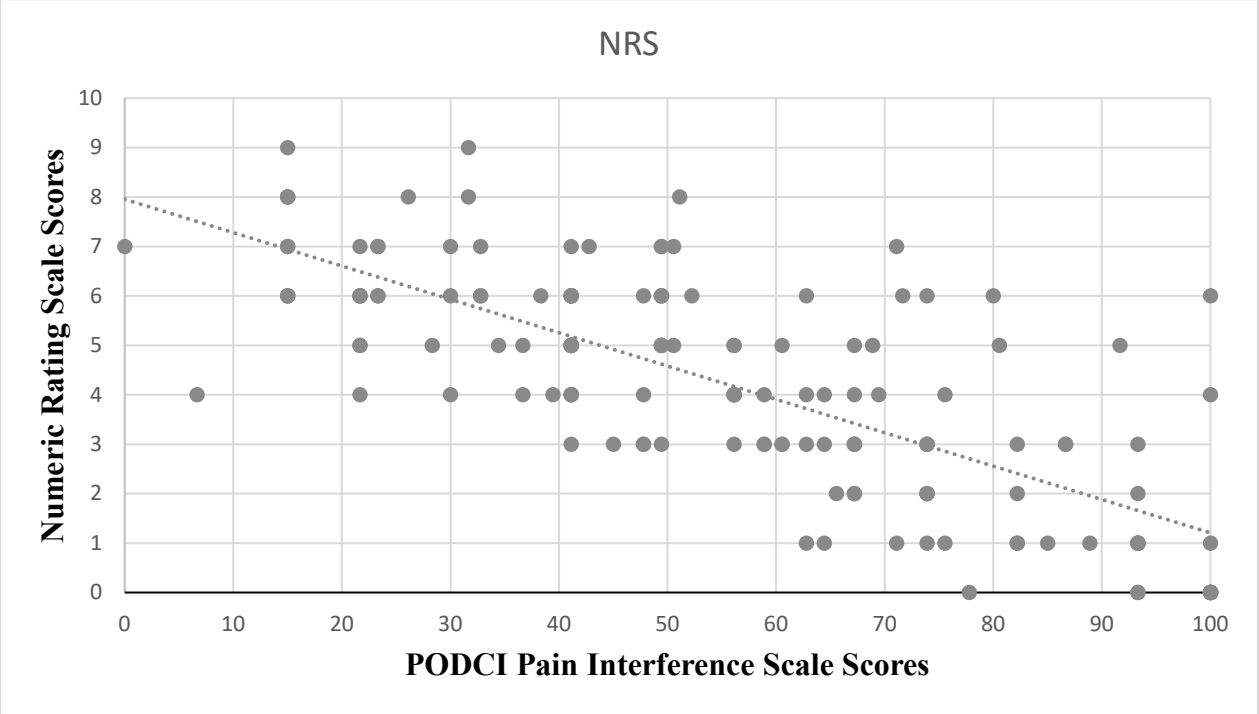
Note. IQR = interquartile range

Figure 1. Scatterplot Showing the Correlation Between the PODCI Upper Extremity Function Scale and the PROMIS Pediatric Upper Extremity Short Form (raw scores)



$r_s = .79, p < .001$

Figure 2. Scatterplot Showing the Correlation between PODCI Pain Scores and Numeric Rating Scale Scores



$r_s = -.73$; $p < .001$

Appendix A

PODCI Upper Extremity Function and Pain Interference Subscales

Q1. During the last week, was it easy or hard for you to lift heavy books?

Easy (1) A little hard (2) Very Hard (3) Can't do at all (4)

Q2. During the last week, was it easy or hard for you to pour a half gallon of milk?

Easy (1) A little hard (2) Very Hard (3) Can't do at all (4)

Q3. During the last week, was it easy or hard for you to open a jar that has been opened before?

Easy (1) A little hard (2) Very Hard (3) Can't do at all (4)

Q4. During the last week, was it easy or hard for you to use a fork and spoon?

Easy (1) A little hard (2) Very Hard (3) Can't do at all (4)

Q5. During the last week, was it easy or hard for you to comb your hair?

Easy (1) A little hard (2) Very Hard (3) Can't do at all (4)

Q6. During the last week, was it easy or hard for you to button buttons?

Easy (1) A little hard (2) Very Hard (3) Can't do at all (4)

Q7. During the last week, was it easy or hard for you to write with a pencil?

Easy (1) A little hard (2) Very Hard (3) Can't do at all (4)

Q8. During the last week, was it easy or hard for you to turn door knobs?

Easy (1) A little hard (2) Very Hard (3) Can't do at all (4)

Q9. In regards to your upper extremity pain: During the last week, how much of the time did pain or discomfort interfere with your activities?

Most of the time Some of the time A little of the time None of the time

Q10. In regards to your upper extremity pain: How much pain have you had during the last week?

None Very mild Mild Moderate Severe Very Severe

Q11. In regards to your upper extremity pain: During the last week, how much did pain interfere with your normal activities (including at home, outside of the home, and at school)?

Not at all A little bit Moderately Quite a bit Extremely

Appendix B

PROMIS Pediatric Item Bank v2.0 – Upper Extremity – Short Form 8a

Pediatric Upper Extremity – Short Form 8a

Please respond to each question or statement by marking one box per row.

	In the past 7 days…	With no trouble	With a little trouble	With some trouble	With a lot of trouble	Not able to do
3860R2r	I could button my shirt or pants	☐ 4	☐ 3	☐ 2	☐ 1	☐ 1
2871R1r	I could open a jar by myself.....	☐ 5	☐ 4	☐ 3	☐ 2	☐ 1
4143R1r	I could open the rings in school binders.....	☐ 5	☐ 4	☐ 3	☐ 2	☐ 1
4112R1r	I could pour a drink from a full pitcher.....	☐ 5	☐ 4	☐ 3	☐ 2	☐ 1
3881R1r	I could pull a shirt on over my head by myself.....	☐ 4	☐ 3	☐ 2	☐ 1	☐ 1
4130R1r	I could pull open heavy doors	☐ 5	☐ 4	☐ 3	☐ 2	☐ 1
2857bR1r	I could put on my shoes by myself.....	☐ 5	☐ 4	☐ 3	☐ 2	☐ 1
4109R1r	I could use a key to unlock a door.....	☐ 5	☐ 4	☐ 3	☐ 2	☐ 1

Last Updated: 28 July 2016

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Appendix C

Numeric Rating Scale (NRS)

Please rate your average pain over the last week on the following scale. 0 means no pain at all and 10 means the worst pain you can imagine.

0 1 2 3 4 5 6 7 8 9 10