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Original article

Validation of the Oswestry Disability Index for pain and disability in arthrogryposis multiplex congenita

Talon Jones [a], Rebecca Miller [a], John T. Street [b], Bonita Sawatzky [b,*]

[a] Faculty of Medicine, University of British Columbia, 317-2194 Health Sciences Mall, Vancouver BC, V6T 1Z3 Canada
[b] Department of Orthopaedics, Faculty of Medicine, University of British Columbia, 818 West 10th Avenue, Vancouver BC, V5Z 1M9 Canada

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ABSTRACT

Objective: Chronic musculoskeletal pain and disability is common in adults with arthrogryposis multiplex congenita (AMC), but validated outcome measures of its related disability are lacking. This study aimed to determine the content and construct validity of the Oswestry Disability Index (ODI) for an AMC-appropriate low-back and lower-extremity pain-related disability questionnaire.
Methods: A mixed methods approach was used to investigate the nature of AMC-related low-back and lower-extremity pain and disability. We included 50 adults with AMC from an international arthrogryposis study. Participants completed 5 pain and disability questionnaires and an interview. Content and construct validity of the ODI in the AMC population was assessed by the proportion of participants who stated ODI domains during the open-ended interview and by R^2 values and Pearson's correlation coefficients (*r*-values), respectively.
Results: The content and construct validity of the ODI were considered moderate to high for measuring low-back pain and lower-extremity disability in the adult AMC population. Participants independently identified many activities of daily living (67%), such as walking, standing, personal care, sitting, lifting and sleeping, already included in the ODI. R^2 values were > 0.25 for all 3 measures, demonstrating the strength of construct validity of the ODI in individuals with AMC.
Conclusion: The ODI is a valid outcome tool for low-back and lower-extremity pain-related disability for patients with AMC. Upper-extremity issues were not addressed by the ODI, which will be further addressed in future research.

1. Introduction

Arthrogryposis multiplex congenita (AMC) is a rare non-progressive condition (1:3000 to 1:5000 live births) [1] involving multiple joint contractures present from birth [2]. Classically, features of AMC include multiple curved fixed joints, muscle atrophy and sometimes additional congenital abnormalities such as pulmonary hypoplasia and micrognathia. Treatment typically involves early correction with physiotherapy and surgery, but once the child outgrows paediatric care, there is a paucity of coordinated care for adults with AMC.

The 2nd International Symposium on Arthrogryposis held in St. Petersburg, Russia [3] highlighted the need for health-related quality-of-life and long-term functional outcomes in adults with AMC. In response, the largest international study involving adults with AMC ($n = 177$) was completed and reported that 75% of participants experienced chronic pain; 88% of participants experienced joint-based pain, primarily in lower extremities, and 49% experienced muscle pain [4]. Because of the multiple etiologies of AMC, the pain experienced is speculated to be nociceptive (both musculoskeletal and visceral) and/or neuropathic [4]. Identifying the pain's origin as nociceptive and/or neuropathic is important because the pain is treated with different classes of medications. Almost all individuals stated that pain medication was required at some time in their lives, and 46% were currently taking regular medication [4]. In a small United Kingdom study of patients with AMC ($n = 96$), half of the participants with pain reported that musculoskeletal pain interrupted daily activities [5].

For the general population, chronic pain impacts physical health, daily activity, psychological health, employment and economic well-being [6]. Chronic pain affects one's ability to perform a range of daily activities; 60% of participants are less able or unable to work outside of the home [7]. The pain is

* Corresponding author. 818 West 10th Avenue, Vancouver BC, V5Z 1M9 Canada.
E-mail addresses: talonj@uvic.ca (T. Jones), rlm793@mail.usask.ca (R. Miller), John.Street@vch.ca (J.T. Street), bonita.sawatzky@ubc.ca (B. Sawatzky).

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multidimensional, affecting an individual's life (general health, employment, disability), thus highlighting the need for individualized and comprehensive management strategies [6], which can lead to an increased positive contribution of individuals to society and less long-term drain on a country's resources [8].

Although chronic musculoskeletal pain is frequently reported by adults with AMC, the impact of this pain experience on individual or societal dimensions of health, disability or quality-of-life has never been analyzed. Additionally, no disease-specific pain or disability questionnaires are available for people with AMC. Existing questionnaires are often specific to other diseases or assume physical characteristics non-inclusive of those with AMC and are thus inappropriate to use and possibly misrepresent the pain-related degree of disability experienced.

Measurement of pain-related disability in adults with AMC could be optimized by use of a disease-specific tool, constructed and validated specifically for adults with AMC. Alternatively, the content and external validation of an existing disability tool in a population of adults with AMC may be considered. For many chronic pain conditions, the impact of pain is measured with generic and disease-specific tools, such as the Oswestry Disability Index (ODI) and the Neuropathic Pain Questionnaire. The ODI is an existing tool originally designed to measure disability due to low-back pain and contains 10 questions designed to assess limitations in various domains of activities of daily living (ADLs), focusing on lower-extremity activities. We chose the ODI for this study because data suggested that the pain-related disability experienced by people with AMC primarily affects the low-back and lower-extremity [4].

The purpose of this study was to validate the ODI as a pain and disability outcome measure in the AMC population. Two hypotheses were investigated:

- the ODI has sufficient content validity to assess low-back and lower-extremity-related disability in patients with AMC;
- the ODI has sufficient construct validity to assess low-back and lower-extremity-related disability in patients with AMC.

2. Methods

2.1. Recruitment

As part of an international quality-of-life study in the AMC population [4], participants were asked if they would like to be contacted for future research studies. Those who responded positively were invited via email to participate in this pain study. Inclusion criteria were a diagnosis of AMC, age $>$ 18 years, ability to speak English, and access to the Internet and telecommunications (telephone or Skype). All participants received an information letter. Consent was implied if they logged into the online system and entered their data. This study was approved by the university and hospital ethics review boards.

2.2. Study design

The study used a mixed methods approach (Consecutive and Simultaneous Qualitative and Quantitative Methods) to investigate the nature, location, frequency, intensity and impact of AMC-related pain and disability. The validity of the ODI was compared to other musculoskeletal, spinal and neurological pain and disability measures [Short-Form McGill Pain Questionnaire, Version 2 (SF-MPQ-2), Brief Pain Inventory (BPI), EuroQol Five Dimensions Questionnaire (EQ-5D) and Medical Outcomes Study Short-Form 36 (SF-36)]. This study received ethical approval from our Institutional Behavioural Research Ethics Board.

2.3. Protocol

Questionnaires: upon expressed interest, participants were sent a direct link to the Rick Hansen Global Research Platform, which provided secure access to the SF-MPQ-2, BPI, ODI, EQ-5D and SF-36.

The SF-MPQ-2 [8] was created by adapting the SF-MPQ pain descriptors. The survey uses a visual analogue and verbal rating scale of pain intensity and 15 pain descriptors rated on a 4-point verbal scale; its reliability and validity has been thoroughly proven. Seven symptoms relevant to neuropathic pain were added to the SF-MPQ-2, and all items use a 0–10 numerical rating scale. Acceptable reliability and validity were proven [8]. The 22 descriptors are categorically arranged as continuous, neuropathic, intermittent or affective pain. The categories are evaluated separately, with scores ranging from 0 to 10, and in one total summative score out of 40.

The Short-Form BPI [9] is a 9-item self-administered questionnaire used to evaluate the severity and impact of pain. It is now widely used in a range of chronic cancer-related and non-malignant pain conditions, including arthritis and low-back pain [10]. Pain severity and interference measured by the BPI are scored separately; our interest was in interference. Severity is scored from 0–40, and interference is normally scored from 0–70.

The ODI [11] was designed for patients with low-back pain and contains 10 sections designed to assess limitations in various ADLs. Each section contains 6 statements, each describing more difficulty in that activity than the preceding statement. The patient selects one statement in each section that most accurately describes their limitation. Many studies have demonstrated the validity and reliability of the ODI for spine-related disabilities [12].

The EQ-5D [13] is a standardized, non-disease-specific instrument used to describe and evaluate health states. It uses a 5-part questionnaire and visual analogue rating scale. Participants rate the impact of their disability on a 5-point scale in terms of mobility, self-care, usual activities, pain/discomfort, and anxiety/depression. The 5 points range from no issues to extreme impact/inability to complete a task. As compared with the SF-36, there is evidence for the construct validity of the EQ-5D, but the SF-36 was shown to be a more sensitive instrument due to its larger number of questions [14].

The SF-36 [15] is a shortened version of its original 149 health status questions and is a general outcome measure using 8 scales to measure 3 aspects of health — functional status, well-being, and "overall evaluation of health". The SF-36 has been shown to be acceptable to patients, internally consistent, and a valid measure of health status [16]. The SF-36 was used in this study as a demographic measure. Each scale is directly transformed into a 0–100 scale, the lower the score, the more disability.

2.4. Interview

Participants completed a 20- to 30-min interview via telephone or Skype to explore the frequency, nature, intensity and impact of their pain and disease-specific impairment. Initial questions were open-ended and inquired the domains of daily living that were most affected by pain. Participants were first asked "What activities of daily living do you find that your pain impacts the most?" Subsequent questions were close-ended and focused on how much pain was experienced for each of the 9 domains of the ODI. Using a 0–10 subjective scoring system, participants were asked to score how much pain they felt while performing the tasks indicated by each ODI domain. "No pain" was assigned the value of 0, and "the worst pain of [their] life" was assigned the value 10. It was made clear to all participants that the question was not asking how much each activity is limited by physical disability, but rather how much pain is experienced.

2.5. Data analysis

To determine whether the ODI had sufficient content validity to assess low-back and lower-extremity pain-related disability in AMC, we calculated the proportion of participants who responded to open-ended questioning with domains already included in the ODI. We also determined the activity domains affected by low-back and lower-extremity pain-related disability that were not addressed by the ODI.

To determine whether the ODI had sufficient construct validity to assess low-back and lower-extremity pain-related disability in AMC, we directly correlated the results of the ODI with those of the SF-MPQ-2 (pain descriptors total score), BPI short-form (pain severity score), and SF-36 (pain sub-score) measures. Correlation analysis involved Pearson's correlation coefficients (r-values) and R^2 values between measured total scores or, in the case of the SF-36, categorical sub-scores. R-values were used to directly describe the relation of the pain as measured by the SF-36 pain sub-score, SF-MPQ and BPI to the mean pain and disability score by the ODI. R^2 values for each relationship were used to explain how the variance in measured disability (measured by the ODI) could potentially be attributed to variance in measured pain (measured by the 3 pain scores).

The SF-36 scores were compared to those obtained in 2017 [4] (by t-test) to ensure that our study accurately reflected the AMC population. Additionally, by using each participant's unique identifier, the 2017 SF-36 data were linked with participants' previously acquired original data to monitor their general health status during the past 12 months for participants included in both studies. In both cases, significance (P-value) was set at 0.05 [4].

3. Results

3.1. Population characteristics

This study included 50 participants (74% female, mean [SD] age 42.38 [13.55] years [range 21–85]). Americans comprised 66% of participants, and the remainders were from the European Union (10%), Canada (8%), Australia (6%), the United Kingdom (4%), New Zealand (4%) and Peru (2%). This demographic information closely matched that of the original 2017 study population (Table 1) [4].

The SF-36 data obtained in this study was compared to the original data to ensure that the health of the participants had been stable in the past year by assessing any change between the 2017 study results and the current results [4]. Direct comparisons of participants' scores between the 2 years showed no significant difference. Additionally, the SF-36 mental component score (MCS) and physical component score (PCS) were calculated to compare the overall mental and physical status of this AMC population to a generalized American population unaffected by disability. The MCS and PCS are scored such that the average unaffected person should score 50 in each domain. The scores obtained for MCS and PCS were 50.63 and 33.21, respectively.

3.2. Pain evaluation measures

The SF-MPQ-2 results (Fig. 1) showed an inverse association between frequency of continuous pain and severity. In terms of continuous aspects of pain, scores for 55% (27) of participants were $\leq 3/10$, and for 45% (23), they were $\geq 4/10$. This inverse linear association was not seen with neuropathic, intermittent or affective pain. Instead, scores for most participants were $\leq 3/10$, with scores for a few individual outliers $\geq 4/10$.

The ODI scores demonstrated that many participants (70.5%) identified their pain-related impairment as 40% or less, placing them into the minimal and moderate disability categories. Scores for severity of pain were $\leq 5/40$ on the BPI with an average BPI score of 14.5 (SD 8.76). The EQ-5D results (Fig. 2) demonstrated that mobility and self-care activities were most impaired, with less severe and/or disabling scores for pain and anxiety/depression.

3.3. ODI content validity

The use of open-ended questioning during the semi-structured interviews determined the domains most commonly affected by pain in this population (Table 2). When self-generated answers were compared to the ADLs included in the ODI, the domains present in both (in decreasing order of prevalence) were walking, standing, personal care, sitting, lifting, and sleeping (Table 3). When questions were phrased in an open-ended manner, no participant identified pain interference in the ODI domains of social activities, travelling or sex life. However, subsequent closed-ended questioning specific to ODI domains showed significant pain-induced impairment in these domains, despite their absence during the initial open-ended inquiry (Table 3). Additionally, several activities not included in the ODI were identified using the open-ended interview questions. Most of these tasks required the use of the upper-extremity, such as writing, typing, driving, household chores, and extracurricular/recreational activities.

3.4. ODI construct validity

The results of the construct validation process are in Table 1. The strongest correlation was found between the SF-36 pain sub-score and the ODI ($r = -0.75$). Similarly, the SF-MPQ2 and BPI scores correlated with the ODI score produced r-values of 0.52 and 0.66, respectively. More importantly, the R^2 values for all 3 pain scores attributed substantial degrees of the variance observed in disability to the variance in pain with this population. When correlated with the ODI, the SF-36 pain sub-score described the highest degree of attribution, with an R^2 value of 0.56. The BPI and SF-MPQ2 followed, with R^2 values of 0.44 and 0.27, respectively.

4. Discussion

In the largest study to date on the long-term outcomes in patients with AMC, axial and appendicular skeletal pain was a

Table 1
Population demographics and outcome measure score correlations with the Oswestry Disability Index (ODI).

	Youngest age, years	Oldest age, years	Mean age (SD), years	Median age (range), years	Male (%)	Female (%)
Current sample	21	85	42.38 (13.55)	41 (21–85)	26	74
2017 study [4]	19	84	39 (NR)	36 (19–84)	29	71

	ODI score, 0–76	SF-36 general health, 5–100	SF-36 pain, 0–100	SF-MPQ-2, 0–28	BPI, 0–33
Mean output score	29.86	59.80	57.70	7.77	12.51
R^2 value		0.41	0.56	0.27	0.44

NR: not reported; SF-36: Medical Outcomes Survey Short-Form 36; SF-MPQ-2: Short-Form McGill Pain Questionnaire, Version 2; BPI: Brief Pain Inventory.

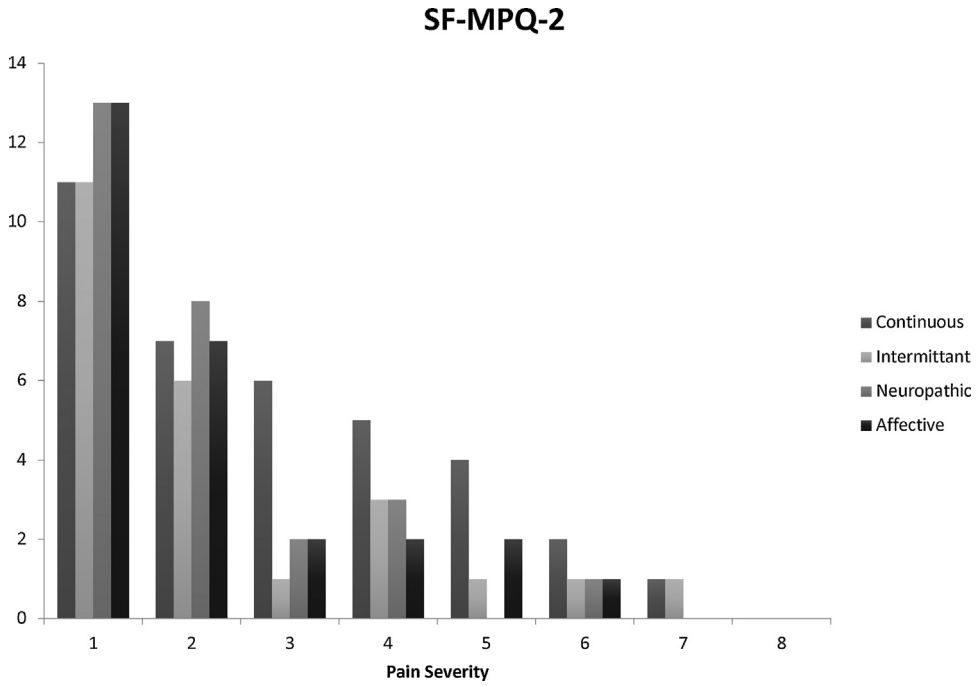

Fig. 1. Association between frequency of continuous pain and severity on the Short-Form McGill Pain Questionnaire, Version 2 (SF-MPQ-2).

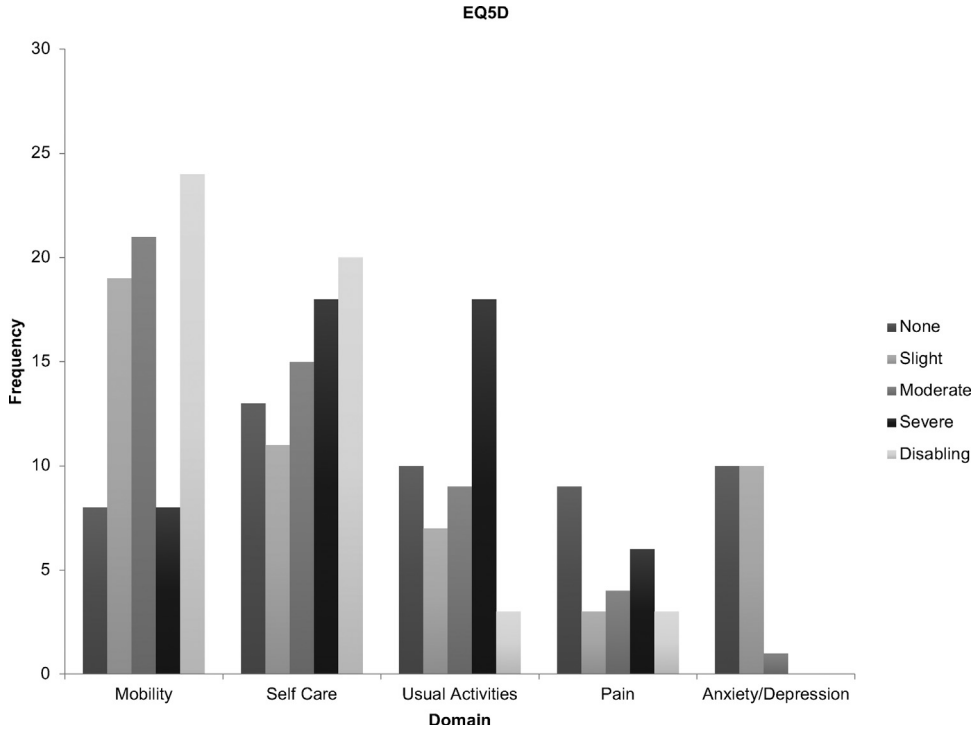

Fig. 2. Degrees of impairment on dimensions of the EuroQol Five Dimensions Questionnaire (EQ-5D).

problem experienced by many participants. Unfortunately, no AMC-specific pain or disability measure is available for follow-up studies in this population. This is the first study to examine the construct and content validity of an existing disability measure commonly used in adult orthopaedics, the Oswestry Disability Index (ODI), to assess low-back and lower-extremity pain-related disability in patients with AMC.

The ODI was designed to assess lower-back pain-related disability and has never been tested in more generalized musculoskeletal conditions such as AMC. We assessed the content validity of the ODI in the AMC population by using open- and close-ended questions of the interview. Participants independently identified many ADLs (67%) included in the ODI. When specifically inquired in a closed-ended format, all ODI domains were shown to affect this AMC population. During open-ended questioning, domains were reported that were not encompassed by the ODI, but these predominantly involved the upper extremities. Therefore, the ODI yielded sufficient content validity for its application in the AMC population in measuring low-back and lower-extremity pain-related disability.

Table 2
Domains affected by pain – frequency of self-generated answers.

Answer provided	Frequency
All tasks	5
Driving	2
Extracurricular/recreational activities	3
Household chores	4
Immobility	4
Lifting	4
Personal care	8
Sitting	7
Sleeping	2
Stairs	8
Standing	15
Swallowing	1
Typing	3
Walking	24
Weather	1
Writing	2
Work	4

Table 3
Percentage of participants who mentioned ODI categories in self-generated answers as compared with mean ODI scores.

ODI category	Percentage of participants (%)	Mean ODI score
Personal care	13.33	2.6
Lifting	6.67	3.1
Walking	40.00	4.3
Sitting	11.67	3.0
Standing	25.00	4.9
Sleeping	3.33	3.8
Sex life	0.00	2.9
Social life	0.00	3.3
Travelling	0.00	3.5

ODI: Oswestry Disability Index.

AMC-related disability is complex and can be a composite of physical limitation, mental health, social barriers, and subjective pain. The intent of the ODI is to measure pain-related disability, specifically of the low-back and lower extremities. To ensure construct validity in the AMC population, it was critical to evaluate how much variance in disability could be attributed to variances in pain. When the scores of each pain measure were plotted against the corresponding participant ODI scores, the R^2 values showed that within this population, the ODI can sufficiently ascribe a great deal of the variance between disability and pain. All 3 measures produced R^2 values > 0.25, which demonstrates the strength of the construct validity of the ODI in individuals with AMC. The results of multiple pain measures (SF-36 pain sub-score, BPI, SF-MPQ-2), when compared to the ODI, supported this association. Our intent was not to compare the efficacy of the pain scores directly but rather to ensure that the ODI redundantly measured disability due to pain regardless of the pain tool applied.

To ensure that our results could be generalized to the AMC population, we compared our sample with the sample in the 2017 study by Nouraei et al. [4]. The 2017 study was the largest international study of AMC and described the most comprehensive use of the SF-36 within the AMC population. This comprehensiveness allowed the findings to be used as a reflection of the generalized, total AMC population. Comparison of the SF-36 scores of this study to the 2017 findings showed no statistically significant difference and, combined with similar MCS and PCS scores, demonstrated that the 2 populations were statistically identical. Our study included 50 participants, which we believe was a significant number due to the disease rarity. Consequently, the results of our study can appropriately be generalized to the total AMC population.

Our study did not classify the participants based on type of AMC, because many of the participants did not have a specific diagnosis beyond that of AMC. However, a recent paper by Dai et al. demonstrated that the prevalence and location of pain did not differ between AMC types [17].

Despite this study's validation of the ODI in the AMC population, several activities not included in the ODI were identified by use of the open-ended interview questions, most involving the use of upper extremities. These activities are not included in the ODI because it is a questionnaire related to low-back pain-related disability. The Dai et al. study found that pain in AMC is predominately located in the trunk and lower limbs and less frequently in the upper extremities [16]. These results support our findings that pain is primarily located in the trunk and lower limbs as opposed to the upper limbs and therefore can be adequately assessed by the ODI. Future steps are to use the Delphi method to establish additional content validity of the ODI. This method will be applied to determine whether the domains not addressed by the ODI are statistically significant within this population. If the domains not addressed are found significant, additional questions will need to be supplemented to and used concurrently with the ODI to adequately assess pain in the AMC population.

5. Conclusions

This study is the first to validate the use of a previously existing tool for assessing pain-related disability in people with AMC. Both the content and construct validity of the ODI were confirmed for low-back and lower-extremity applications, and the results obtained can be generalized to the worldwide AMC population. Despite this validation, there are aspects of pain in AMC that are not addressed by the ODI because of its focus on the low-back and lower-extremity, and the next step will be to use the Delphi method to identify these domains and determine whether they are statistically significant.

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Disclosure of interest

The authors declare that they have no competing interest.

Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at https://doi.org/10.1016/j.rehab.2018.05.1319.

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