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# Psychometric properties of the brief pain inventory modified for proxy report of pain interference in children with cerebral palsy with and without cognitive impairment

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## Abstract

**Introduction:** Cerebral palsy (CP) is the most common cause of physical disability in children and is often associated with secondary musculoskeletal pain. Cerebral palsy is a heterogeneous condition with wide variability in motor and cognitive capacities. Although pain scales exist, there remains a need for a validated chronic pain assessment tool with high clinical utility for use across such a heterogeneous patient population with and without cognitive impairment.

**Objectives:** The purpose of this study was an initial assessment of several psychometric properties of the 12-item modified brief pain inventory (BPI) pain interference subscale as a proxy-report tool in a heterogeneous sample of children with CP with and without cognitive impairment.

**Methods:** Participants (n = 167; 47% male; mean age = 9.1 years) had pain assessments completed through caregiver report in clinic before spasticity treatment (for a subgroup, the modified BPI was repeated after procedure). To measure concurrent validity, we obtained pain intensity ratings (Numeric Rating Scale of pain) and pain intensity, duration, and frequency scores (Dalhousie Pain Interview).

**Results:** Modified BPI scores were internally consistent (Cronbach $\alpha$ = 0.96) and correlated significantly with Numeric Rating Scale intensity scores ($rs$ = 0.67, $P$ < 0.001), Dalhousie Pain Interview pain intensity ($rs$ = 0.65, $P$ < 0.001), pain frequency ($rs$ = 0.56, $P$ = 0.02), and pain duration scores ($rs$ = 0.42, $P$ = 0.006). Modified BPI scores also significantly decreased after spasticity treatment (pretest [scored 0–10; 3.27 ± 2.84], posttest [2.27 ± 2.68]; $t$ (26) = 2.14, 95% confidence interval [0.04–1.95], $P$ = 0.04).

**Conclusion:** Overall, the modified BPI produced scores with strong internal consistency and that had concurrent validity as a proxy-report tool for children with CP.

**Keywords:** Cerebral palsy, Children, Pain interference, Pain intensity, Brief pain inventory, Numeric Rating Scale, Dalhousie Pain Interview

## 1. Introduction

Cerebral palsy (CP) is the most common cause of physical disability in children, affecting 2 to 2.5 of every 1000 live births.[20] Cerebral palsy can result in impaired motor development, muscle stiffness, asymmetric gross motor function, and/or spasticity,[20] and often life-long secondary musculoskeletal pain.[18] Recurrent pain is common and severe; for example, in a large registry study, 37% of children with CP reported pain.[26] Pain seems to be more prevalent (50%–54%) in children with the greatest gross motor impairments.[1,26]

Although there is little debate that individuals with CP experience a great deal of pain, routine and thorough clinical assessment of pain remains a challenge. Multiple pain assessments have been developed that are very appropriate for pain assessment in some individuals with CP. For example, the Non-Communicating Children's Pain Checklist–Revised (NCCPC-R)

or the Paediatric Pain Profile are useful for patients who are nonverbal with or without cognitive impairments or intellectual disability.[7,13] The PROMIS pain interference assessment may be useful for individuals with CP who are able to walk and run.[25] However, to date, no single pain assessment has been established for use across variable motor and cognitive ability levels in CP. This poses a clinical challenge because medical providers and their support staff must become familiar with and adopt multiple pain assessments and become skillful at matching the correct assessment to each individual patient. This creates undue burden during the clinical implementation of chronic pain assessment practices.

The Initiative on Methods, Measurement, and Pain Assessment in Clinical Trials (IMMPACT) consensus panel recommended that the impact of pain on functioning (interference) be included as an outcome in all chronic pain trials.[23] The 7-item pain interference subscale of the brief pain inventory (BPI) was specifically highlighted as a recommended option for assessment of pain-related functional impairment.[9] The original BPI is a 2-part assessment of pain intensity and pain interference with activities of daily living (ADLs). The BPI was originally developed to assess cancer-related pain in adults.[8] Since then, the BPI has also shown strong test–retest reliability and validity in noncancer-related acute and chronic pain in adulthood.[3,22] For example, the BPI showed high internal consistency between the Intensity scale ($\alpha = 0.85$) and the Interference scale ($\alpha = 0.88$) in veterans with chronic, intractable pain.[22] How well the BPI performs as a self-report tool for typically developing children and adolescents is not as well understood. Batalha and Mota[2] used the BPI as a self-report measure to assess the efficacy of massage for cancer pain in typically developing children and adolescents.

Tyler et al.[24] first modified the BPI pain interference subscale for use as a stand-alone self-report pain assessment in a sample of 50 verbal adults with CP without cognitive impairment. The subscale was modified from the original 7 items to include 3 additional pain interference items (ie, pain interference with "self-care," "recreational activities," and "social activities") and to account for participants who were unable to walk because of disability.[24] Specifically, pain interference with "walking ability" was changed to "mobility (ability to get around)." In this first application of the modified BPI, the total interference score correlated significantly with average pain intensity and produced strong internal consistency coefficients. Engel et al.[10] further modified the BPI pain interference subscale for youth with neuromuscular disease, creating a 12-item measure. In this version, "normal work" was changed to "school/work" to accommodate younger participants, and pain interference with "communication with others" and "learning new information or skills" was added. In Engel's study, both self and caregiver report was collected. Parents reported 30 of the children (71%) had chronic pain, whereas 23 (55%) of the youths themselves reported having chronic pain. Only the youth's scores on the modified BPI were reported.

In our previous use of the 12-item–modified BPI pain interference subscale, in caregivers of 34 children with CP, the assessment was easy to administer and seemed widely useful as a proxy-report tool for caregivers of children with CP with cognitive impairments.[1] Pain duration was positively correlated with the total pain interference subscale score.[1] The modified BPI showed promise as a clinically feasible proxy-report measure. The findings have not been replicated with larger samples to further test the validity of the approach in diverse populations with CP and especially in those with limited ability to self-report because of motor or cognitive impairment.

The purpose of the current study was to extend the investigation of the 12-item–modified BPI pain interference subscale for clinical use across patient groups with CP. Specifically, we examined the psychometric properties of the modified BPI for use as a caregiver proxy-report tool to measure pain interference in children with CP with various levels of motor and cognitive impairments. The study aimed to examine the internal consistency of BPI item scores, as well as concurrent validity with pain intensity, duration, and frequency scores generated using a Numeric Rating Scale (NRS) of pain and the Dalhousie Pain Interview (DPI) by interviewing caregivers of children with CP.

## 2. Methods

### 2.1. Brief overview: two data sets

Data reported here were drawn from 2 separate studies assessing pain in patients with CP associated with spasticity. Data from the first study (study 1) were collected as part of a study assessing residual procedural pain in the 2 days after botulinum toxin injections. In study 1, the modified BPI was used along with a NRS of pain. Data from the second study (study 2) were collected as part of an ongoing study investigating pain outcomes after intrathecal baclofen (ITB) pump implant. In study 2, the modified BPI was used along with the DPI, which obtains a thorough assessment of pain type, pain intensity using a NRS of pain, pain frequency, and pain duration. In both study 1 and study 2, all baseline pain information, including pain intensity and pain interference, were collected from caregivers (proxy report). In addition, a convenience subgroup of caregivers in study 2 completed the modified BPI again after ITB pump implant (allowing for a preliminary test of sensitivity to change in pain and pain interference).

### 2.2. Participants and setting

Participants were enrolled in study 1 if they were attending a midwestern tertiary children's specialty hospital for spasticity management (ie, botulinum toxin injections), had a diagnosis of CP, read and spoke the English language (or family members did), and had a caregiver present to complete the study tasks. Because of the low risk and clinical nature of study 1, a passive consent process was approved by the ethics review board, whereby verbal and written language informed caregivers that if they completed the brief questionnaire, they were consenting to participate in the study. In study 2, participants were enrolled if they were attending a midwestern tertiary children's specialty hospital for spasticity management (ie, ITB pump implant), had a diagnosis of CP or related developmental disorder associated with spasticity, were between 3 and 40 years of age, spoke the English language (or family members did), had a caregiver present to complete the study tasks, provided assent/consent, and did not have a comorbid psychiatric disorder (eg, major depression) or a co-occurring chronic pain condition (eg, juvenile rheumatoid arthritis).

Participants, combined from studies 1 and 2, included 167 individuals with CP (47% male; mean chronological age = 9 years, 2 months; range 2 months–34 years, 6 months). Demographic information included specific CP diagnosis, Gross Motor Function Classification System (GMFCS) level, ethnicity, age, verbal ability, and level of cognitive impairment (**Table 1**). Most participants were pediatric (<18 years, n = 163; 98%) and had a CP diagnosis of quadriplegia (n = 105; 63%). Most

**Table 1**

**Participant demographics.**

| | Total sample (n = 167) | Study 1 NRS (n = 89) | Study 2 DPI (n = 78) |
|---|---|---|---|
| Mean age (y) | 9.18 (5.13) | 7.93 (4.29) | 10.61 (5.64) |
| Age range | 2 mo–34 y | 2 mo–17 y | 4–34 y |
| Male, n (%) | 79 (47.3) | 48 (53.9) | 31 (39.7) |
| Ethnicity | | | |
| White | 129 (77.2) | 64 (71.9) | 65 (83.3) |
| African American | 15 (9.0) | 11 (12.4) | 4 (5.1) |
| Asian | 3 (1.8) | 2 (2.2) | 1 (1.3) |
| Native American | 2 (1.2) | 0 (0) | 2 (2.6) |
| Hispanic/Latino | 5 (3.0) | 4 (4.5) | 1 (1.3) |
| Jamaican | 1 (0.6) | 0 (0) | 1 (1.3) |
| Multiple ethnicities | 11 (6.6) | 8 (9.0) | 3 (3.8) |
| Unreported | 1 (0.6) | 0 (0) | 1 (1.3) |
| CP diagnosis, n (%) | | | |
| Hemiplegia | 32 (19.2) | 31 (34.8) | 1 (1.3) |
| Diplegia | 27 (16.2) | 11 (12.4) | 16 (20.5) |
| Triplegia | 9 (5.4) | 6 (6.7) | 3 (3.8) |
| Quadriplegia | 97 (58.1) | 39 (43.8) | 58 (74.4) |
| Unreported | 2 (1.2) | 2 (2.2) | 0 (0) |
| GMFCS level, n (%) | | | |
| Level 1 | 29 (17.4) | 24 (27.0) | 5 (6.4) |
| Level 2 | 18 (10.8) | 14 (15.7) | 4 (5.1) |
| Level 3 | 18 (10.8) | 8 (9.0) | 10 (12.8) |
| Level 4 | 35 (21.0) | 19 (21.3) | 16 (20.5) |
| Level 5 | 57 (34.1) | 14 (15.7) | 43 (55.1) |
| Unreported | 10 (6.0) | 10 (11.2) | 0 (0) |
| Verbal ability | | | |
| Verbal | 84 (50.3) | 43 (48.3) | 41 (52.6) |
| Nonverbal | 53 (31.7) | 16 (18.0) | 37 (47.4) |
| Unreported | 30 (18.0) | 30 (33.7) | 0 (0) |
| Cognitive impairment | | | |
| None | 31 (18.6) | 19 (21.3) | 12 (15.4) |
| Mild | 30 (18.0) | 15 (16.9) | 15 (19.2) |
| Moderate | 20 (12.0) | 11 (12.4) | 9 (11.5) |
| Severe | 37 (22.2) | 10 (11.2) | 27 (34.6) |
| Unreported | 49 (29.3) | 34 (38.2) | 15 (19.2) |
| Pain present previous week | 91 (54.5) | 25 (28.1) | 66 (84.6) |

CP, cerebral palsy; DPI, Dalhousie Pain Interview; GMFCS, Gross Motor Function Classification System; NRS, Numeric Rating Scale.

caregivers (n = 91; 54%) endorsed that their child was currently experiencing pain or had experienced pain in the week prior. Approval for the study was given through the University of Minnesota Institutional Review Board, and all participants gave informed consent in accordance with the Declaration of Helsinki.

### 2.3. Study 1 procedures

In study 1, caregivers completed the modified BPI and a NRS of pain while in the clinic room waiting for their child to receive botulinum toxin injections. Data were collected as part of a research study measuring residual procedure-related pain in the 2 days after botulinum toxin injections. Caregivers completed the modified BPI to assess pain interference with 12 ADLs in the previous 7-day period. Caregivers also completed a 11-point NRS (n = 89) to assess pain intensity "right now" (before receiving injections). Participant demographic information was collected from the medical record.

### 2.4. Study 2 procedures

In study 2, caregivers completed the modified BPI and the DPI pain assessments in the presurgical clinic room while waiting for their child to receive an ITB pump implant. Data were collected as part of

an ongoing research study measuring pain and spasticity outcomes after ITB pump implant. The DPI (n = 78) was completed through caregiver interview to document pain intensity, frequency, and duration for each type of pain reported.[1,5] For 27 participants in study 2, the BPI and DPI were reassessed 6 months after ITB pump implant. Intrathecal baclofen pump treatment would be expected to decrease spasticity and associated musculoskeletal pain. Demographic information, cognitive impairment, verbal ability, motor function, and pain type, and location were collected through direct caregiver interview.

### 2.5. Measurement of pain interference

The modified BPI was used as a measure of pain interference with ADLs. The specific modifications to the BPI pain interference items were those created by Engel et al.[10] for use in individuals with neuromuscular disease. The modified BPI version used included assessment of pain's interference with 12 ADLs over the course of the previous week.[10] Activities included general activity, mood, mobility (ability to get around), school, work or other chores, relations with other people, sleep, enjoyment of life, self-care (taking care of your daily needs), recreational activities, social activities, communication with others, and learning new information. Each item was scored on a 0 to 10 scale (0 = pain

does not interfere; 10 = pain completely interferes). Total scores were calculated by totaling each item's score, with total scores ranging from 0 to 120. Alternatively, total scores can be divided by 12 to provide scores in the 0 to 10 range, which may be more clinically intuitive. Brief pain inventory scores have previously correlated with pain intensity and have demonstrated excellent internal consistency (Cronbach α ≥ 0.89) in adults with CP[24] and in a small sample of children with CP.[1]

### 2.6. Measurement of convergent pain constructs

In study 1, pain intensity was assessed by asking caregivers to rate how strong their child's pain was "right now," before receiving botulinum toxin injections. Responses were documented independently in paper format using a 11-point NRS with zero indicating "no pain at all" and 10 indicating "worst pain ever." The NRS of pain was completed concurrently with the BPI. The NRS is among the most frequently used pain assessment measures in research and clinical care with adequate reliability and validity in experimental conditions.[12,14,27] The NRS has also been shown to be highly sensitive to changes in pain scores and is recommended for clinical and research purposes.[11]

In study 2, the DPI was conducted in an interview/script format consisting of 2 sections: recent pain experience (1-week recall of any pain events is documented in detail) and chronic pain (pain lasting longer than 6 months[16]). This format was adapted from previous studies using proxy caregiver report when self-report is not possible.[5] The DPI assessed pain intensity in the previous week using an 11-point NRS from 0 to 10 (0 = no pain at all; 10 = worst pain ever), pain frequency (estimate number of pain episodes in previous week), and pain duration (estimate total amount of time with pain in the previous week) for each type of pain reported by the caregiver (eg, headache pain and spasm pain). Each pain type was recorded and categorized as accidental, gastrointestinal, musculoskeletal, neurological, stretching, positioning, equipment, orthopedic, spasm, other, or unknown pain. The most common pain types endorsed in the study 2 sample included musculoskeletal-, spasm-, and gastrointestinal-related pain.

### 2.7. Measurement of participant descriptors

Demographic information, including specific CP diagnosis, GMFCS level, cognitive impairment level, and verbal ability, were collected from the medical record when available in provider dictation notes. In study 2, there was an opportunity to obtain additional information from caregivers related to these participant descriptors. Motor impairment was assessed using the GMFCS,

which can validly and reliably classify individuals with CP.[4] The GMFCS rating scale consisted of I to V levels of gross motor function based on functional limitations, with level I indicating the highest gross motor function and level V indicating the lowest function.[28] Gross Motor Function Classification System rating was determined through medical records or using functional information provided by caregiver proxy report if GMFCS information was not available in the medical record.[15]

### 2.7.1. Statistics

Normality of data was estimated using the Shapiro–Wilk test. Internal consistency was assessed using Cronbach α. Spearman correlation coefficient (rs) for nonnormally distributed data was used to assess the relationship between pain interference (BPI) and other pain parameters (NRS and DPI). Additional correlations were conducted with 2 subgroups of children based on cognitive impairment level: (1) children with no impairment or mild cognitive impairment, and (2) children with moderate to severe cognitive impairment. Dalhousie Pain Interview subscales used in analysis included pain intensity, pain frequency, and pain duration. Pain parameters for the DPI are summed to include data for all pain types/locations reported. Because multiple types of pain often coexist, the summary data can exceed the 0 to 10 scale or the number of hours in a week. Significance was set at $P < 0.05$ for all tests. To test GMFCS level as a potential predictor of modified BPI score, a simple linear regression model was developed and fitted separately. All statistical tests were completed using SPSS version 20 (SPSS Inc, Chicago, IL).

## 3. Results

### 3.1. Pain experience

For study 1, in which participants were seen before botulinum toxin injections, 28% of caregivers reported that their child with CP was experiencing pain "right now." Numeric Rating Scale ratings of pain were on average 1.04 of 10 (median = 0.00; range 0–9). For study 2, in which participants were seen before an ITB pump implant, 77% of caregivers reported that their child had experienced pain in the previous week. Pain intensity, duration, and frequency of episodes are displayed by pain type in **Table 2**.

### 3.2. Brief pain inventory validity evidence and sensitivity

The modified BPI demonstrated excellent internal consistency (Cronbach α = 0.96, n = 167). All corrected item total correlations for the 12 items were ≥0.77. There were 33 instances where the modified BPI detected pain interference in

**Table 2**

**Pain parameters by pain type for study 2 (n = 78).**

| Pain | MSK | Stretching | Spasm | GI | Accidental | Headache | Other |
|---|---|---|---|---|---|---|---|
| Incidence n (%) | 37 (47.44) | 22 (28.21) | 14 (17.95) | 11 (14.10) | 6 (7.69) | 5 (6.41) | 4 (5.13) |
| Constant pain n (%) | 14 (17.95) | 0 (0) | 1 (1.28) | 0 (0) | 0 (0) | 0 (0) | 1 (1.28) |
| Intensity, M (SD) Range | 5.54 (2.31) 1–10 | 4.36 (1.76) 1–10 | 5.86 (2.14) 2–10 | 5.92 (2.10) 2–9 | 4.17 (1.50) 1–6 | 4.33 (1.56) 2–6 | 4.25 (2.88) 1–10 |
| Duration (h), M (SD) Range (h) | 66.80 (78.71) 0.0008–168 | 0.06 (0.09) 0.0008–0.47 | 18.38 (30.75) 0.004–168 | 9.96 (11.72) 0.05–48 | 3.02 (4.99) 0.001–18 | 1.27 (0.86) 0.08–3 | 43.19 (62.41) 0.33–168 |
| Pain episodes, M (SD) Range | 11.41 (12.14) 1–70 | 7.14 (3.91) 1–21 | 22.93 (28.17) 1–210 | 9.33 (9.22) 1–60 | 1 (0) 1–1 | 1 (0) 1–1 | 2.50 (1.50) 1–5 |

GI, gastrointestinal-related pain; M, mean; MSK, musculoskeletal pain; Other, other types of pain including teeth pain (n = 2), menstrual pain (n = 1), and pain related to having the common cold (n = 1); Spasm, spasm-related pain.

**Table 3**

**Brief pain inventory (BPI) degree of pain interference with activities of daily living (ADLs).**

|  | Interference severity, n (%) | | Interference tally, n (%) | |
|---|---|---|---|---|
|  | Low (score ≤ 4) | High (score ≥ 5) | Absent (score = 0) | Present (score ≥ 1) |
| General activity | 143 (85.6) | 24 (14.4) | 93 (55.7) | 74 (44.3) |
| Mood | 130 (77.8) | 37 (22.2) | 88 (52.7) | 79 (47.3) |
| Mobility | 129 (77.2) | 38 (22.8) | 85 (50.9) | 82 (49.1) |
| School, work, other chores | 136 (81.4) | 31 (18.6) | 102 (61.1) | 65 (38.9) |
| Relations with other people | 148 (88.6) | 19 (11.4) | 112 (67.1) | 55 (32.9) |
| Sleep | 135 (80.8) | 32 (19.2) | 99 (59.3) | 68 (40.7) |
| Enjoyment of life | 140 (83.8) | 27 (16.2) | 98 (58.7) | 69 (41.3) |
| Self-care | 145 (86.8) | 22 (13.2) | 117 (70.1) | 50 (29.9) |
| Recreational activities | 143 (85.6) | 24 (14.4) | 106 (63.5) | 61 (36.5) |
| Social activities | 147 (88.0) | 20 (12.0) | 116 (69.5) | 51 (30.5) |
| Communication with others | 150 (89.8) | 17 (10.2) | 119 (71.3) | 48 (28.7) |
| Learning new information | 147 (88.0) | 20 (12.0) | 120 (71.9) | 47 (28.1) |

the previous week that caregivers did not report using the NRS of pain "right now" or the DPI (previous week). Conversely, there were 6 instances where caregivers reported pain experience using the NRS or DPI but did not report pain's interference with ADLs on the BPI. Despite these differences, there was a statistically significant association ($\chi^2$ (1) = 49.91, $P < 0.001$) between assessments for the detection of pain presence vs absence using the BPI and NRS or DPI.

Overall, BPI scores indicated that participants experienced pain in the previous week that interfered with ADLs (ADL; **Table 3**). Mean pain interference was greatest for mood (2.01 ± 2.76) and mobility (2.12 ± 2.92), and least for communication with others (1.14 ± 2.31). Overall, 52 (31.1%) participants experienced substantial pain interference (rated ≥ 5 on the 0–10 scale) with at least 1 ADL. The ADLs most frequently endorsed as substantially impacted by pain (scored ≥ 5/10) in this sample included mood (22.2% of respondents), mobility (22.8% of respondents), school, work or other chores (18.6% of respondents), and sleep (19.2% of respondents; **Table 3**).

Gross Motor Function Classification System level significantly predicted BPI pain interference scores $r^2 = 0.11$ (adjusted $r^2 = 0.10$; β = 0.33, $t$ (155) = 4.37, 95% confidence interval [CI] [3.17–8.39], $P < 0.001$) but not NRS pain intensity ($r^2 = 0.02$; adjusted $r^2 = 0.01$; β = 0.15, $t$ (77) = 1.33, 95% CI [−0.10 to 0.51], $P = 0.19$), DPI pain intensity ($r^2 = 0.002$; adjusted $r^2 = -0.011$; β = −0.047, $t$ (76) = 0.408, 95% CI [−0.85 to 1.29], $P = 0.69$), pain duration ($r^2 = 0.04$; adjusted $r^2 = 0.03$; β = 0.20, $t$ (76) = 1.79, 95% CI [−4630.86 to 86,626.58], $P = 0.078$), or frequency of pain episodes ($r^2 = 0.04$; adjusted $r^2 = 0.03$; β = 0.20, $t$ (76) = 1.78, 95% CI [−0.56 to 10.06], $P = 0.08$).

**Table 4**

**Brief pain inventory (BPI) total and item scores and correlations with pain intensity.**

| BPI component | Item score, mean (SD) | Correlation to NRS ($r_s$) | | |
|---|---|---|---|---|
|  |  | Total sample (n = 89) | No–mild CI (n = 34) | Mod–sev CI (n = 21) |
| Total BPI | 1.54 (2.18) | 0.672* | 0.642* | 0.829* |
| General activity | 1.47 (2.35) | 0.590* | 0.501* | 0.758* |
| Mood | 2.01 (2.76) | 0.548* | 0.411† | 0.727* |
| Mobility | 2.12 (2.92) | 0.545* | 0.433† | 0.537† |
| School, work, other chores | 1.69 (2.68) | 0.588* | 0.598* | 0.422 |
| Relations with other people | 1.17 (2.12) | 0.592* | 0.427† | 0.583* |
| Sleep | 1.86 (2.92) | 0.611* | 0.654* | 0.587* |
| Enjoyment of life | 1.63 (2.55) | 0.581* | 0.492* | 0.713* |
| Self-care | 1.29 (2.57) | 0.532* | 0.575* | N/A |
| Recreational activities | 1.53 (2.70) | 0.579* | 0.416† | 0.701* |
| Social activities | 1.30 (2.48) | 0.591* | 0.488* | 0.828* |
| Communication with others | 1.14 (2.31) | 0.573* | 0.519* | 0.635* |
| Learning new information | 1.21 (2.52) | 0.592* | 0.594* | 0.532† |

\* Significant at the $P < 0.001$ level.
† Significant at $P < 0.05$ level.
BPI, brief pain inventory; CI, cognitive impairment; Mod, moderate; N/A, caregivers reported this item as either "0" (pain did not interfere) or as "not applicable" to their child; NRS, Numeric Rating Scale; $r_s$, Spearman correlation coefficient; Sev, severe.

### 3.3. Study 1: association between Numeric Rating Scale of pain intensity and brief pain inventory pain interference

The modified BPI total score displayed a strong, significant correlation with the NRS pain intensity score ($rs = 0.67$, $P < 0.001$; **Fig. 1**). As observed for all 12 interference items and the average pain intensity score, the correlation coefficients were significant at the $P < 0.001$ level with a coefficient range of 0.53 to 0.67. These results held when subgroup analyses were conducted by cognitive impairment status. For children with no impairment or mild cognitive impairment, the BPI total score correlated with the NRS pain intensity score ($rs = 0.64$, $P < 0.001$, n = 34). For children with moderate to severe cognitive impairments, the BPI correlated with the NRS pain intensity score to an even greater degree ($rs = 0.83$, $P < 0.001$, n = 21). See **Table 4** for all item-level correlations.

### 3.4. Study 2: association between Dalhousie Pain Interview items and brief pain inventory pain interference

Brief pain inventory total score significantly correlated with DPI pain intensity ($rs = 0.65$, $P < 0.001$; **Fig. 2**), pain frequency ($rs = 0.56$, $P < 0.001$), and pain duration ($rs = 0.42$, $P < 0.001$); however, correlations with pain frequency and duration seemed to be influenced by outliers. Brief pain inventory scores significantly decreased after putative pain/spasticity treatment (ITB implant pretest [scored 0–10; 3.27 ± 2.84], posttest [2.27 ± 2.68]; $t$ (26) = 2.14, 95% CI [0.04–1.95], $P = 0.04$). When subgroup analyses were conducted for study 2, the sample size of children with no impairment or mild cognitive impairment was small (n = 12) and likely underpowered for the correlational analyses between the BPI score and DPI pain intensity ($rs = 0.20$, $P = 0.53$), pain duration ($rs = 0.38$, $P = 0.22$), or pain frequency ($rs = 0.35$, $P = 0.27$). For children with moderate to severe cognitive impairments, the BPI correlated significantly with DPI pain intensity ($rs = 0.71$, $P < 0.001$, n = 21) and duration ($rs = 0.47$, $P = 0.01$), but not pain frequency ($rs = 0.32$, $P = 0.10$).

## 4. Discussion

In response to the IMMPACT consensus panel recommendation that the interference items of the BPI be used to assess pain-related functional impairment,[9] we tested several psychometric properties of a modified BPI for use as a caregiver proxy-report pain interference tool in a heterogeneous clinical sample of patients with CP. The sample was inclusive of a wide range of ages (albeit mostly children and youth), cognitive abilities, motor function, and CP types. There were a number of specific findings. Importantly, the modified BPI seemed to produce reliable and valid scores reflecting pain's impact on individuals with CP. In some instances, the BPI was more likely to detect the presence of pain (or perhaps more accurately, its effect on function) compared with the DPI or the NRS of pain "right now." The latter is especially important because the NRS of pain "right now" is often used by providers to determine whether pain is an issue to be addressed during a medical visit. In addition, BPI scores significantly decreased after a putative spasticity and musculoskeletal pain intervention (ITB pump implant). To our knowledge, this is the first use of the BPI to prospectively document ITB pain–related outcomes in CP. Finally, GMFCS level significantly predicted BPI total score, suggesting that individuals with the least ability to be mobile are at greatest risk for pain interference with ADLs.

Over half of the sample (54%) had experienced pain in the previous week, and nearly a third of the sample (31%) had pain that severely impacted at least one activity of daily living (considered scores ≥5 of 10 on BPI item). Overall, pain seemed to be poorly managed. Breau et al.[6] first noted that pain experience has a significant impact on persons', with intellectual and developmental disabilities, ability to function. Similarly, caregivers in this study noted a high degree of pain interference with ADL. On average, pain most impacted mobility, mood, sleep, school/work/chores (adapted from "normal work"), and enjoyment of life (in that order). Pain's interference with these ADL is similar to a previous study in neuromuscular disease where pain most commonly impacted sleep, general activity, mood, mobility, and school/work/chores.[10] Typically developing chronic pain populations often report similar pain interference profiles with these same activities being most impacted.[8,17,21,22] For example, Osborne et al.[17] found that among patients with multiple sclerosis, sleep, recreational activities, enjoyment of life, normal work, and mobility were the highest ranked areas of pain impact.
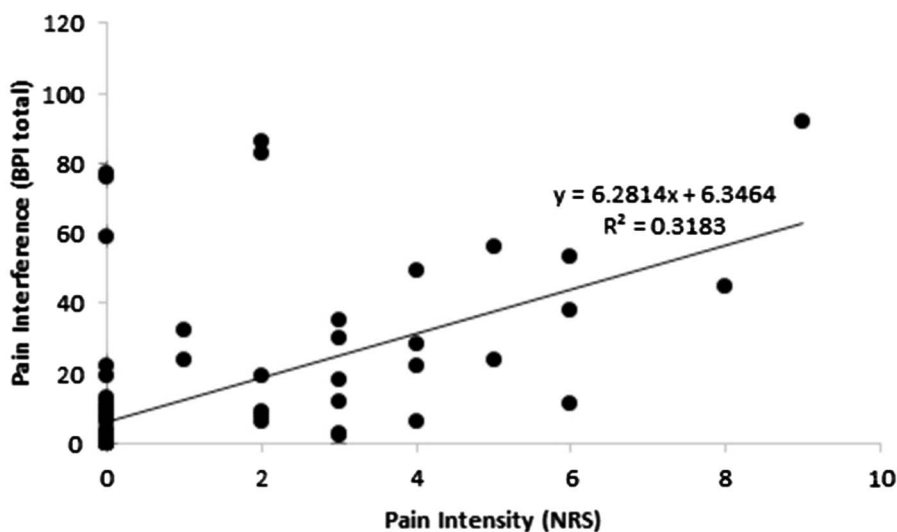


**Figure 1.** Relationship between the modified brief pain inventory (BPI) pain interference subscale score (scored 0–120) and the Numeric Rating Scale (NRS) of pain intensity score (scored 0–10).
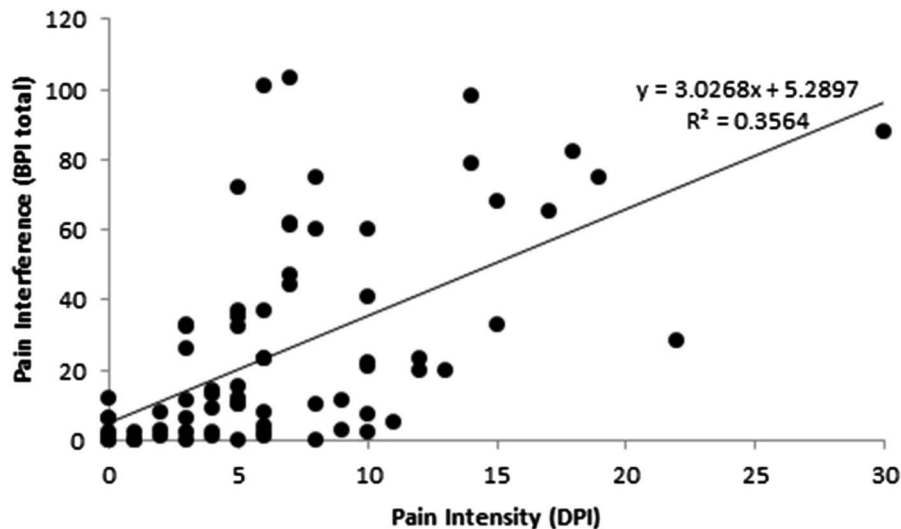
**Figure 2.** Relationship between the modified brief pain inventory (BPI) pain interference subscale score (scored 0–120) and the Dalhousie Pain Interview (DPI) pain intensity measure (each pain type is scored 0–10 then summed for a total score).

Given the IMMPACT recommendations,[9] along with the psychometric properties of the BPI interference items provided here and in previous studies, it seems reasonable to recommend the BPI pain interference items for use in future research studies and in further testing for application in clinical practice as an appropriate means to assess pain-related functional impairment in CP. The relatively large data set presented with broad age range, mobility levels, CP types, and cognitive abilities suggest that the modified BPI is appropriate for most persons with CP. Although our sample had few adult patients relative to pediatric (children and youth), Tyler et al.[24] previously reported on the strong psychometric properties of the BPI interference items for use as a self-report measure for adults with CP. Here, we report on similar psychometric properties of the modified BPI as a proxy-report tool for ages 2 months through adulthood. The modified BPI may be among the first pain assessment tools to show promise across both types of assessment (self and proxy) with broad ranges of ages and abilities in CP. Future research is needed to explore the modified BPI as a child self-report tool for those with CP with no cognitive impairment or mild cognitive impairment.

There were study limitations that should be noted. First, data were collected as part of 2 distinct ongoing research protocols. As such, the NRS rating scale in study 1 asked about pain "right now" rather than pain in the previous 7 days. This may account for the comparatively low pain intensity scores in study 1; however, a correlation between pain "right now" and pain interference in the previous week was statistically significant. Second, although both subsamples included participants with a primary diagnosis of CP, there were clearly differences between the groups on severity of CP and CP type. We consider this a possible strength, however, in terms of creating a sample that is representative of the population of individuals with CP. That said, the sample was one of clinical convenience sampling and was not formed by random sample, and therefore, the results are specific to the sample, and any inferences to the CP population should be performed with appropriate caution. Finally, we relied on proxy report for all participants. One next step might include a study focused on assessing the utility of the modified BPI for use as a self-report tool in adolescents and older children with CP without cognitive impairments. This would determine whether the tool is feasible for use in that age/ability group.

## 5. Conclusions

Based on the current findings, the modified BPI may be considered for use as proxy-report tool to assess pain interference with ADL in CP across a wide range of ages and developmental and physical abilities. This study adds to the work demonstrating strong psychometric properties of the modified BPI for use as a self-report tool in adults with no cognitive impairments as well as across ages and developmental disabilities as a proxy-report tool in CP.[1,19,24] The modified BPI is recommended for use in clinical research to further establish feasibility for caregivers and patients/participants to complete quickly and with little to no support from staff.

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