Parent described pain responses in nonverbal children with intellectual disability

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A R T I C L E   I N F O

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A B S T R A C T

Background: Assessing pain for nonverbal children with intellectual disability is challenging. These children are at risk for pain from complex medical conditions and the interventions to treat those conditions. Pain responses are often misunderstood, given that the children are nonverbal and limited by their physical abilities.

Objectives: The general purpose was to examine the words that parents of children with intellectual disability use to describe their child's pain response in order to improve pain recognition and management. Specifically, the aims were to: (1) identify common pain responses as reported by parents; (2) examine relationships between type of pain response and independent variables; (3) compare pain responses to those identified in the literature.

Design: The study design includes: (1) a summative qualitative content analysis of the parent described pain responses; (2) quantitative analysis of the relationships between type of pain response and independent variables and; (3) a comparison of pain responses to those identified in the literature.

Setting: A pediatric hospital in the northeastern United States.

Participants: Parents of 50 nonverbal children with intellectual disabilities ages 6–18 years.

Methods: A non-directed summative content analysis identified patterns within 335 parent described pain responses. Chi square analysis was used to examine relationships between type of pain response and selected demographic factors.

Results: Seven distinct categories of pain responses were identified. The most commonly reported pain responses were within the categories of: vocalization (39.4%), social behavior (21.8%) and facial expressions (16%). Four types of pain responses: vocalization, social behavior, muscle tone and activity level included opposite responses to pain. There were significant relationships between type of pain response and (1) pain severity; (2) causes of intellectual disabilities and; (3) sex of the child. Pain responses varied with severity. Patients with seizure disorders expressed pain more vocally. Females expressed pain with more social responses and males with more vocalizations.

Conclusions: The results support evidence that parents can articulate their child’s pain responses. The study provides evidence of: (1) extremes of pain responses and (2) a significant relationship between type of responses and severity of pain, cause of intellectual disability and sex of child.

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What is already known about the topic?

- Nonverbal children with intellectual disability are at risk for having pain.
- Pain assessment in nonverbal children with intellectual disability is challenging.
- Difficulties in pain recognition can have serious consequences.

What this paper adds

- Evidence that parents can articulate their child’s pain responses.
- Evidence of extremes of pain responses in this population.
- Documentation of a significant relationship between type of responses and severity of pain, cause of intellectual disability and sex of child.

1. Background

Pain is a subjective experience best understood through self-reported descriptions using words and phrases. When self-reports of pain are not possible, as is the case with nonverbal children with intellectual disability, assessment of behavior and other physical changes are the next best sources toward understanding their pain experiences. Pain recognition is crucial to identifying the source of pain, treating health issues and providing safe, effective pain management especially in nonverbal children with intellectual disabilities who often have complex medical conditions (Stallard et al., 2001). In this population responses to pain have been described as: (1) individual (McGrath et al., 1998; Terstegen et al., 2003; Carter et al., 2002); (2) idiosyncratic (Fanurik et al., 1999); and (3) dampened and delayed (Oberlander et al., 1999). A better understanding of pain expressions and the relationships between these expressions and child characteristics (cause of intellectual disability, co-morbidities, pain severity, developmental age, communication ability, socialization sex of child) may help us to differentiate how certain children express pain.

Children with intellectual disability make up a small percentage of the general public, 0.3–2.5% of the United States population, depending on how intellectual disability is defined (United States Department of Education Statistics). However, these children are at risk for having health issues (van Schrojenstein Lantman-De Valk et al., 2000; Gibert-MacLeod et al., 2000) and pain (Stallard et al., 2001; Hadden and von Baeyer, 2002). In a prospective study of nonverbal children with intellectual disabilities, 73.5% of the children (n=25) experienced pain at least one day during a 2-week period as assessed and recorded by their parents. Parents assessed moderate to severe pain in 68% of their children (Stallard et al., 2001).

Even with an obvious source, such as surgical pain, children with intellectual disabilities may not receive the interventions that others receive. In a study comparing the administration of opioids 1–3 days after a spinal fusion, children with intellectual disabilities received smaller total doses of opioids than those without intellectual disabilities (Malviya et al., 2001). One reason for the under treatment is that pain in this population may not be easily recognized. Another reason may be that the amount of opioids estimated by clinicians to be safely administered may be reduced because of concerns of opioid related respiratory depression due to the child’s co-morbidities.

Difficulties in pain recognition can have serious consequences. A study of deaths caused by intestinal obstructions found an unusually high mortality rate due to nonmalignant gastrointestinal obstruction in individuals with intellectual disabilities (Jancar and Speller, 1994). Some have interpreted these tragic cases as a sign of pain indifference or pain insensitivity (Biersdorf, 1994, 1991). However, other studies have found that although pain responses may be subtle or different, pain responses in children with intellectual disabilities are identifiable especially by parents and caregivers (Fanurik et al., 1999; Hunt et al., 2003; McGrath et al., 1998; Terstegen et al., 2003). The high mortality rates may be related to delays in pain recognition, which delay evaluation of medically fragile people.

Several teams of researchers have begun to describe how children with intellectual disabilities respond and react to pain. There are 6 pain assessment tools published in the English Language with psychometric testing: Face-Legs-Activity-Cry and Consolability (Merkel et al., 1997); Individualized Numeric Rating Scale (Solodiuk and Curley, 2003); Non-communicating Children’s Pain Checklist (McGrath et al., 1998; Breau et al., 2000, 2001, 2002a,b; Kleinnekhe, 2007; Johansson and Carlberg, 2010); Pain Indicator for Communicatively Impaired Children (Hunt et al., 2004, 2007); Pediatric Pain Profile (Stallard et al., 2002); University of Wisconsin Children’s Hospital Pain Scale (Soetenga et al., 1999). Despite 6 published pain assessment tools for this population, at this time, there is still no consensus about how to best assess pain in this population. The lack of consensus persists because of the difficulties capturing the varied pain expressions described in this population. A description of each of the above mentioned pain assessment tools is outlined in Section 2.

The general purpose of this study was to examine the words that parents of children with intellectual disability use to describe their child’s pain response in order to improve pain recognition and management. Specifically, the aims were to: (1) identify common pain responses as reported by parents; (2) examine relationships between type of pain response and independent variables; (3) compare pain responses to those identified in pain assessment tools described in the literature.

2. Methods
2.1. Conceptual framework

This study is grounded in the Social Communications Model of Pain. This model integrates the biological, psychological and social aspects of pain. The model describes how a pain stimulus is processed by the individual and how observers decode the pain expressions of another (Craig, 2009). In nonverbal children, decoding pain expressions is the basis for all pain assessment. In this
study, relationships were measured between the child’s pain responses and biology (sex, cause of intellectual disability, co-morbidities, pain severity); psychology (developmental age) and the social aspects of pain (communication ability, socialization) (Fig. 1).

2.2. Sample

With approval from the hospital’s institutional review board and parental consent, data on a prospective, convenience sampling of 50 parents of nonverbal children with intellectual disabilities (ages 6–18) was collected from the preoperative and orthopedic clinics at a 350 bed acute care pediatric hospital in the northeastern United States. Data was collected for this study between July 2004 and April 2008 during a primary study measuring the psychometric properties of the Individualized Numeric Rating Scale (Solodiuk et al., 2010). The inclusion criteria for both studies were: (1) age 6–18 years with intellectual disabilities; (2) the above described child was scheduled for elective surgery requiring an overnight stay in the hospital. Exclusion criteria were: (1) the above described child could communicate verbally even with one word or via a communication board and (2) the child was not currently living with their families, or had not lived with their families in the last 6 months.

2.3. Procedures

Potential subjects were screened and informed of the study by a preoperative or orthopedic clinic nurse. The principal investigator or a research assistant then discussed the study and obtained consent. The preoperative clinic nurse asked the parent to describe their child’s responses when at their baseline and not having acute pain and when they knew the child had pain. Parents were asked to link the responses with a number from 0 to 10 to indicate pain severity. For example, parents may report their child has no pain when giggling and babbling; pain of 5/10 when crying out; pain of 8/10 when crying with tears and; pain of 10/10 when their child stops vocalizing and closes his eyes. These responses were used to populate the Individualized Numeric Rating Scale. The responses that parents linked to no pain were used by clinicians as a baseline to compare other behaviors to.

After the child’s surgery, during subsequent meetings to collect data for the primary study, the principal investigator or research assistant asked parents to review the populated Individual Numeric Rating Scale and either to confirm or make any changes to the tool in order to verify and if necessary, clarify the collected data on the Individual Numeric Rating Scale. Data collection included simultaneous but independent pain assessments by parents, bedside nurses and the principal investigator or research assistant.

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Fig. 1. Model of the integration of the biological, psychological, social aspects of pain in nonverbal children with intellectual disability. Adapted from the Social communication Model of Pain (Craig, 2009; Hadjistavropulos & Craig, 2002).
2.4. Measures

2.4.1. Face-Legs-Activity-Cry and Consolability (FLACC)

The FLACC is a 5 item behavioral pain assessment tool originally created for children without intellectual disabilities (Merkel et al., 1997), but validated in children with intellectual disabilities (Malviya et al., 2006; Voepel-Lewis et al., 2002). In the first study of the FLACC for children with intellectual disabilities, the tool was not modified (Voepel-Lewis et al., 2002). The FLACC scores by nurses correlated with parent scores \( (p < 0.001) \). The FLACC scores decreased after administration of analgesics \( (p < 0.001) \). The best correlation in scores was in categories of Face and Cry \( (r = 0.505–0.698, k = 0.33–0.448 \) and \( r = 0.638–0.826, k = 0.434–0.652 \). Measures of exact agreement were acceptable for most categories; however, the category of least agreement was in the Legs category \( (17–88\%, k = 0.205–0.477) \) (Voepel-Lewis et al., 2002). Although the study showed good validity and reliability, the sample chosen for this study had a wide range of intellectual disabilities. Forty-nine percent \( (n = 39) \) of children had some verbal ability and 30% \( (n = 24) \) had good verbal communication. Only 51% \( (n = 40) \) were not able to communicate verbally. Likely, this sample with a wide range of verbal abilities was chosen in an effort to demonstrate the generalizability of the tool for a wide range of children. Further research is needed to determine if the same results would be found in a sample of nonverbal children with intellectual disabilities.

In 2005, the FLACC was revised to include allow for individualization. Initial findings support the reliability and validity of the revised FLACC as a measure of pain in children with intellectual disabilities \( (intra-class correlation coefficient \( [ICC] = 0.87–0.92; \) criterion validity \( r = 0.65–0.87; \) construct validity using paired \( t \) tests decrease in pre to post analgesic pain scores \( (p < 0.001) \) \) (Malviya et al., 2006). However, the study sample also had a wide variety of intellectual disabilities \( 46\% (n = 24) \) with significant intellectual disabilities; 23% \( (n = 12) \) had moderate impairment and 31% \( (n = 16) \) had mild impairment. So it is unknown whether the positive results were influenced by assessing children with a high level of expressive abilities.

2.4.2. Individualized Numeric Rating Scale

The Individual Numeric Rating Scale is a numeric rating scale with space to populate the child’s behavior when in pain and not (Soloduk and Curley, 2003). Inter rater reliability was tested across triads of raters (parent, bedside nurse and research nurse) before \( (n = 85) \) and after \( (n = 85) \) an intervention for pain. Inter rater agreement between the parents and research nurse was high \( (ICC = 0.82–0.87) \) across all ratings. Parent and bedside nurse agreement for ratings immediately before a pain intervention \( (ICC = 0.65) \) and post intervention \( (ICC = 0.74) \) also suggested good reliability. Agreement between bedside nurse and research nurse \( (ICC = 0.74–0.80) \) also suggest good reliability. The study also reported moderate to strong correlation between Individual Numeric Rating Scale and Non-communicating Children’s Pain Checklist-Post Operative Version \( (0.63–0.73) \) (Soloduk et al., 2010).

Further research is needed to measure results in different clinical settings and types of pain.

2.4.3. Non-communicating Children’s Pain Checklist

The Non-communicating Children’s Pain Checklist is the most studied pain assessment tool for this population (McGrath et al., 1998; Breau et al., 2000, 2001, 2002a,b; Kleinknecht, 2007; Johansson and Carlberg, 2010). This 31 item checklist was originally created from the descriptions of pain responses reported in interviews of 20 parents or caregivers of children with intellectual disability (McGrath et al., 1998). A revised 30 item version of the checklist was tested in the home setting with parents or caregivers reporting whether the items were present or not present in 4 situations: (1) an acute episode; (2) when long term pain was present; (3) during a non-painful but distressing situation and; (4) when the child was calm (Breau et al., 2000). More pain responses were present during pain when compared to a calm situation. The responses did not differentiate between pain and a no-pain but distressed situation.

The later version of this checklist, the Non-communicating Children’s Pain Checklist-Postoperative Version contains 27 items classified into six subscales (Breau et al., 2002b). This revised tool was tested in 24 children as observed by parent or caregiver and researchers in a hospital setting. The tool was internally consistent \( (Cronbach’s alpha of caregivers scores = 0.91 \) and Cronbach’s alpha of researchers scores = 0.71 \) with good inter rater reliability in some of the categories post operatively \( (ICC = 0.77 \) for vocal subscale, 0.48 for social, 0.81 for facial, 0.61 for activity, 0.45 for body and limb, and 0.63 for physiologic) (Breau et al., 2002b). There were significant correlations between most of the caregiver and researcher scores after surgery \( (0.64–0.72) \). However, the correlation between the caregivers and researchers scores before surgery, were not significant. Also, there were no significant correlations between the proxy visual analog scale scores completed by nurses and the Non-communicating Children’s Pain Checklist-Post Operative Version scores completed by parents and caregivers (Breau et al., 2002b). This is an important consideration because patients in a hospital setting are frequently assessed for pain by nurses. Further investigation is needed to evaluate clinical utility.

2.4.4. Pain Indicator for Communicatively Impaired Children

The Pain Indicator for Communicatively Impaired Children is a pain assessment tool created for children with intellectual disabilities (Stallard et al., 2002). Six pain responses for this population were identified: “crying with or without tears”; “screaming, yelling, groaning or moaning”; “screwed up or distressed looking face”; “body appears stiff or tense”; “difficult to comfort or console”; “flinches or moves away if touched”. Using these 6 pain responses, parents assessed their child’s pain at 14 different times over a one-week period of time. Parents also documented the presence or absence of pain. One item, “screwed up or distressed looking face” correctly predicted 87% of pain in 49 children as assessed by their parents or caregivers (Stallard et al., 2002). Further testing
is needed to assess whether the tool would be accurate when used by caregivers without knowledge of the child’s pain responses.

2.4.5. Pediatric Pain Profile

The Pediatric Pain Profile is a 20 item pain assessment tool created for children with intellectual disabilities (Hunt et al., 2004, 2007). It was developed through parent interviews of their child’s pain responses for children with intellectual disabilities who were unable to communicate through speech or augumentative communication. Inter rater reliability testing of the Pediatric Pain Profile was completed by parent assessments and simultaneously by a co-rater identified by the parents, some of whom were health care providers (Hunt et al., 2004). Inter rater reliability ranged from an ICC of 0.74–0.89. Further testing is needed by caregivers without knowledge of the child’s pain responses.

2.4.6. University of Wisconsin Children’s Hospital Pain Scale

The University of Wisconsin Children’s Hospital Pain Scale is an assessment tool created for preverbal and nonverbal children (Soetenga et al., 1999). Initial testing of the scale showed good validity (internal consistency = 0.93) and reliability (inter rater reliability = 0.92). However, the small number of nonverbal children with intellectual disabilities (n = 15) which was combined with preverbal children (n = 59) makes it difficult to evaluate its usefulness for children with intellectual disabilities.

2.4.7. Vineland Adaptive Behavior Scale

The child’s level of functioning was measured by the Vineland Adaptive Behavioral Scale through semi structured interviews of the parents. The Vineland Adaptive Behavioral Scale has 301 items listed developmentally with likert responses to measure the child’s level of functioning in 4 domains: communication, daily living skills, socialization and motor skills. The psychometrics of the Vineland Adaptive Behavioral Scale are internal consistency (split-half 0.76–0.99), with test retest reliability of 0.76–0.93, and inter rater reliability of 0.62–0.78 (Rosenbaum et al., 1995; Vig and Jedrysek, 1995). Validity was measured with a high correlation (0.65–0.94) between Vineland II (Sattler and Hoge, 2006) and the Vineland Adaptive Behavioral Scale. Results of the Vineland Adaptive Behavioral Scale are reported in months.

2.5. Data analysis

The first step in the analysis was a summative qualitative content analysis conducted by the principal investigator. This is the process of interpreting, classifying and comparing content into themes and patterns with counts of words and phrases (Hsieh and Shannon, 2005). This technique has been used to evaluate topics such as the content of nursing textbooks related to death and dying (Ferrell et al., 1999) and the content of lumbar spine X-ray reports (Thompson and Carr, 2007). To prepare for the data analysis, the handwritten, populated Individual Numeric Rating Scales were transcribed into spreadsheets and cleaned. Statistical Package for Social Sciences (SPSS) version 15.0 (IBM, Somers, NY) was used because the number of subjects (n = 50) and parent described pain responses (n = 423) were not easily accommodated by usual qualitative methods of data analysis.

The analysis was non-directed, meaning that the categories for evaluating the frequency counts were not predetermined, but were established during the analysis. Categories of similar pain responses were clustered together. For example, social behaviors as pain responses were grouped together, such as “withdraws from others” and “tries to hide and bury self”. Throughout this process of preliminary organizing, the data were examined for the emergence of new categories and categories were readjusted and defined. Most of the pain responses fit easily into categories. A 3 member panel of experts reviewed the categories of pain responses and provided judgments on ambiguous pain responses that did not fit easily into one category. The expert panel consisted of nurses with experience in post-operative pain in children with intellectual disabilities. The panel included 3 surgical nurses (2 bedside nurses on a surgical unit, 1 master prepared surgical nurse practitioner). The panel was chosen for their interest in pain management and this population. The panel’s mean years of nursing experience was 28 years with a range from 26 to 35 years. The expert panel was white (100%), female (100%). To avoid premature analytic closure due to an a priori view (Sandelowski, 1995), nurses from the expert panel were not involved in the primary study. Once the final categories were derived and defined, frequency counts were calculated.

The next step in the analysis required quantitative methods. The relationship between categories of pain response (nominal level data) as determined by the qualitative analysis and the following independent variables was examined using chi square statistics. The independent variables collected as nominal data were sex of child and reason for intellectual disabilities. The independent variables collected as interval data (child expressiveness, child developmental age, severity of pain and child co-morbidities) were re-grouped as nominal data. For example, severity of pain was re-grouped from interval (0–10) to categorical data: no pain (0), mild pain (1–3), moderate pain (4–7), and severe pain (8–10). Individually, contingency tables were set up for each of the pain responses (vocalization-yes, vocalization-no) and the each of the independent variables.

The final step was a comparison of the parent described pain responses with those identified in the literature in the 5 previously described standardized pain assessment tools. The tools included in this analysis were: r-FLACC, Non-communicating Children’s Pain Checklist, Pain Indicator for Communicatively Impaired Children, Pediatric Pain Profile and University of Wisconsin Children’s Hospital Pain Scale. The Individual Numeric Rating Scale was not evaluated in this analysis because the parent described pain responses used in this study were obtained from this tool. In order to examine the comprehensiveness of the pain assessment tools, the 335 parent described pain
responses were re-categorized according to the items described in pain assessment tools.

3. Results

3.1. Child demographics

The demographics of the convenience sample of 50 nonverbal children ages 6–18 years with intellectual disabilities are listed in Table 1. The most common cause of intellectual disabilities was peri-natal events \( (n = 30) \). The cause of impairment was unknown for nine children and information was unavailable for two. Other causes of intellectual disability included epileptic syndromes \( (n = 4) \); autism \( (n = 2) \) and; chromosomal syndromes \( (n = 3) \). The children were admitted for the following surgeries: orthopedic surgeries other than spinal fusions \( (n = 26, 52\%) \); spinal fusion \( (n = 16, 32\%) \); general surgeries \( (n = 6, 12\%) \); otolaryngology surgeries \( (n = 2, 4\%) \).

3.2. Parent demographics

The parents of the previously described children were mostly female \( (88\%) \), white \( (80\%) \), with a mean age of 40 years \( \text{range} 27–59 \text{ years old} \) (Table 2).

3.2.1. Results Aim #1: categorized parent described pain responses

All 50 parents were able to describe and rank order their child’s pain responses and link them to numbers indicating pain intensity. The mean number of descriptors per parent \( (8.4 \text{ descriptors}) \) and the distribution of the descriptors by severity on a scale from 0 to 10 are described in Figs. 1 and 2. Parents reported the majority \( (53.4\%) \) of their child’s pain responses at 3 points on the 0–10 continuum: no pain

![Figure 2](https://via.placeholder.com/150)

Fig. 2. Number of parent described pain responses per patient.
(n = 88, 20.8%); at a pain intensity of 5 (n = 57, 13.5%) and a pain intensity of 10 (n = 76, 18%) (Fig. 2).

Of the total 423 parent described pain responses, parents linked 88 to when the child had no pain or was in her usual state. Although baseline behaviors are important to use for comparison when assessing for pain, they were not included in the analysis of pain responses since they do not describe a pain state. It is important to note that 25% (n = 22) of the no pain descriptors (n = 88) such as “not vocalizing”, “active, moves a lot”, “seeks physical closeness”, “moans” or “sleeps” were responses to pain in other children.

Most of the remaining 335 pain responses fit easily into 7 distinct categories: (1) vocalizations including crying; (2) social behaviors; (3) facial expression; (4) physiologic measures; (5) muscle tone; (6) activity level and; (7) self-injurious behaviors. The greatest percentage of pain responses were clustered within the categories of vocalizations including crying (39.4%), followed by facial expression (21.8%) and social behaviors (16%) (Table 3). Within four of the categories, vocalization, social behaviors, muscle tone and activity level, the pain responses had a wide range of responses, and even included disparate responses to pain. For example, some children had increased vocalizations while others decreased vocalizations in response to pain. Some responded to pain by withdrawing while others reached out for human comfort. Most responded to pain with increased muscle tone while others had decreased muscle tone. Some children exhibited increased activity while others exhibited decreased activity level when in pain.

3.2.2. Results Aim #2: relationship between pain response and other variables

The relationships between the categories of parent described pain responses (7 point nominal data as listed in Table 3) and certain independent variables (listed in Table 4) were each explored individually using chi square analyses (Table 4). Severity of pain was significantly related to the categories of pain responses $[\chi^2 (21, n = 423) = 451.13, p < .001]$. Parents reported facial responses to pain more often when describing their child’s reaction to mild and moderate rather than severe pain. Incrementally, the number of parent described pain responses within the category of vocalization increased with pain severity (Fig. 3). Physiologic responses to pain were described more often during moderate to severe pain. Self-injurious behavior and social behavior did not increase as pain severity increased.

Causes of intellectual disabilities were collected by reviewing the medical record and clarifying this information with the parent. Cause of intellectual disabilities was significantly related to the pain categories $[\chi^2 (14, n = 423) = 38.79, p < .000]$ with more changes in vocalizations and more self-injurious behaviors being reported in children with seizure disorders (Fig. 4). The relationship between the sex of the child and categories of pain response was significant $[\chi^2 (7, n = 423) = 18.62, p < .01]$. Parent’s reported more physiologic and social responses in daughters and more changes in vocalization and fewer physiologic and social changes in

<table>
<thead>
<tr>
<th>Table 3</th>
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<tr>
<td>Categorized pain responses: frequencies and examples.</td>
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<table>
<thead>
<tr>
<th>Categories</th>
<th>n</th>
<th>%</th>
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<tbody>
<tr>
<td>Vocalizations including crying</td>
<td>132</td>
<td>39.4%</td>
</tr>
<tr>
<td>Social behaviors</td>
<td>73</td>
<td>21.8%</td>
</tr>
<tr>
<td>Facial expression</td>
<td>53</td>
<td>16.0%</td>
</tr>
<tr>
<td>Physiologic</td>
<td>24</td>
<td>7.2%</td>
</tr>
<tr>
<td>Muscle tone</td>
<td>23</td>
<td>6.9%</td>
</tr>
<tr>
<td>Activity level</td>
<td>20</td>
<td>6.0%</td>
</tr>
<tr>
<td>Self-injurious behaviors</td>
<td>9</td>
<td>2.7%</td>
</tr>
<tr>
<td>Total</td>
<td>335</td>
<td>100%</td>
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<th>Table 4</th>
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<tr>
<td>Chi square analysis: pain response and independent variables (n = 423).</td>
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<table>
<thead>
<tr>
<th>Independent variables</th>
<th>df</th>
<th>$\chi^2$</th>
<th>p value</th>
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<tbody>
<tr>
<td>Child characteristics</td>
<td></td>
<td></td>
<td></td>
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<tr>
<td>Severity of pain</td>
<td>21</td>
<td>451.13</td>
<td>$p &lt; .001^*$</td>
</tr>
<tr>
<td>Causes of ID</td>
<td>14</td>
<td>38.79</td>
<td>$p &lt; .000^*$</td>
</tr>
<tr>
<td>Child sex</td>
<td>7</td>
<td>18.62</td>
<td>$p &lt; .01$</td>
</tr>
<tr>
<td>Child co-morbidities</td>
<td>14</td>
<td>20.46</td>
<td>0.12</td>
</tr>
<tr>
<td>Child communication ability</td>
<td>153</td>
<td>140</td>
<td>0.21</td>
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<tr>
<td>Child socialization</td>
<td>14</td>
<td>8.45</td>
<td>0.87</td>
</tr>
<tr>
<td>Child daily living skills</td>
<td>14</td>
<td>13.92</td>
<td>0.46</td>
</tr>
<tr>
<td>Child developmental age</td>
<td>14</td>
<td>19.156</td>
<td>0.16</td>
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<tr>
<td>Parent characteristics</td>
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<tr>
<td>Parent sex</td>
<td>7</td>
<td>7.8</td>
<td>0.35</td>
</tr>
<tr>
<td>Parent age</td>
<td>21</td>
<td>23.18</td>
<td>0.34</td>
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df: degrees of freedom; $\chi^2$: chi square statistics; ID: intellectual disability.

* Significance.
sons. The number of the child's co-morbidities, sex of parent, parent age, 3 domains of the Vineland Adaptive Behavioral Scale: child communication ability, daily living skills and social skills and child developmental age (total score on the Vineland Adaptive Behavioral Scale) were not significant in this analysis. Motor skills, the 4th domain of the Vineland Adaptive Behavioral Scale, were not assessed in this population as the participants were extremely limited in their independent motor skills. The additional questions for motor skills assessment were considered burdensome to parents without being of benefit to the study.

3.2.3. Results

Aim #3: comparison of pain responses to published pain assessment tools

Each of the 5 pain assessment tools in this analysis had outliers of the 335 pain responses that were not able to be categorized into the items described by the pain tools. For example, parent described pain responses that were physiological (such as increased, heart rate, increased blood pressure, sweating, seizure activity) could not be categorized into the items described in the r-FLACC. Of the 5 tools examined, the Non-communicating Children’s Pain Checklist was the most comprehensive. Only 2.8% (n = 12) of the 335 pain responses were not included within the tool's items. Both the University of Wisconsin Children’s Hospital Pain Scale and the Pediatric Pain Profile were the least comprehensive with 31.9% (n = 135) of the 335 parent described pain responses not included within the items of those pain assessment tools (Table 5).

4. Discussion

The results of this study are congruent with the Social Communication Model of Pain. These results support the concept that decoding pain responses is a complex interaction between the person experiencing pain and observers attempting to understand the experience. This
study supports evidence that parents of children with intellectual disabilities are able to decode and articulate their child’s pain responses (McGrath et al., 1998; Terstegen et al., 2003; Fanurik et al., 1999; Hunt et al., 2003; Carter et al., 2002). Parental descriptions of pain responses are the foundation for most pain assessment tools for this population either during tool development (McGrath et al., 1998; Hunt et al., 2004, 2007) or individualization (Malviya et al., 2006; Solodiuk et al., 2010). In addition, parent-proxy ratings in nonverbal children with intellectual disability correlated well with nurse-proxy ratings (ICC = 0.78 [confidence interval = 0.63–0.87] and ICC = 0.73 [confidence interval 0.59–0.83]) respectively (Voepel-Lewis et al., 2005).

Carter et al. (2002) described the process of how parents of children with intellectual disability learn their child’s pain responses in a qualitative case study design. Parents (n = 15) described 3 processes used in assessing pain: “guessing about the pain, working it out and instinctively or intuitively knowing” (Carter et al., 2002, p. 452). “Guessing” about the pain is used when parents begin to learn pain responses. Guessing is based on knowledge of the child and the situation. “Working it out” describes a systematic approach of knowing the child’s common ailments, intervening and watching for the response. The last process, “instinctively knowing” described a process in which parents internalize the process so that they identify pain responses and possible causes quickly. Throughout this study, most parents populated the Individual Numeric Rating Scale with speed and confidence. However, one parent of an autistic boy had more difficulty articulating her son’s pain behaviors and she listed fewer pain descriptors. Her son was included in the study because he met the criteria of being nonverbal with intellectual disability. This parent reported that her son did not always respond consistently to similar painful stimuli such as receiving immunizations at the pediatrician’s office. Further research is needed to understand the

<table>
<thead>
<tr>
<th>Tool</th>
<th># Items</th>
<th>Not included</th>
<th>Examples of responses not included</th>
<th>Tool’s lowest frequency item</th>
<th>Tool’s highest frequency item</th>
</tr>
</thead>
<tbody>
<tr>
<td>r-FLACC</td>
<td>5</td>
<td>17% (n = 71)</td>
<td>Seizure activity</td>
<td>“Legs” 0.5% (n = 2)</td>
<td>“Crying” 27.9% (n = 118)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td>(includes all vocalizations-can individualize)</td>
<td></td>
</tr>
<tr>
<td>NCCPC</td>
<td>30</td>
<td>2.8% (n = 12)</td>
<td>Heart rate high</td>
<td>“Shivering” 0% (n = 0)</td>
<td>“Cry” 15.8% (n = 67)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td>(separate category for “special sound or vocalization”)</td>
<td></td>
</tr>
<tr>
<td>PICIC</td>
<td>6</td>
<td>19.9% (n = 84)</td>
<td>Floppy arms</td>
<td>“Flinches or moves away when touched” 4.7% (n = 20)</td>
<td>“Crying” 17% (n = 72)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>PPP</td>
<td>20</td>
<td>31.9% (n = 135)</td>
<td>Sweats</td>
<td>“Flexed inward or drew legs upwards toward chest” 0.2% (n = 1)</td>
<td>“Crying, moaned, groaned” 26.7% (n = 113)</td>
</tr>
<tr>
<td></td>
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<td></td>
<td></td>
</tr>
<tr>
<td>UWCH</td>
<td>5</td>
<td>31.9% (n = 135)</td>
<td>Arms floppy</td>
<td>“Sleep” 0.5% (n = 2)</td>
<td>“Vocal/Cry” 23.2% (n = 98)</td>
</tr>
<tr>
<td></td>
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<td></td>
</tr>
</tbody>
</table>
child, parent and interpersonal elements that may condense or prolong this process of how parents learn about their child’s responses to pain.

The results of the categorized parent described pain responses and frequency counts supports the Social Communication Model of Pain. In this study, the majority of pain responses were grouped into 3 categories: (1) vocalization including crying (39.4%); (2) social behaviors (21.8%) and: (3) and facial expression (16%). The primary function of these 3 categories of human response is communication. In contrast, the primary functions of the other 4 categories of pain response (physiologic indicators, muscle tone, activity level, and self-injurious behaviors) are not commonly ascribed to communication. Physiologic responses to pain comprised only 7.2% of the pain responses reported by parents. It is possible that physiologic responses to pain were not commonly described by parents because parents do not readily recognize physiologic changes. Parents may not have easy access to physiologic responses to pain such as heart rate, blood pressure, oxygen saturation. Another explanation is that the low percentage of physiologic pain descriptors reported by parents may be because this population often has pain daily (Stallard et al., 2001; Hadden and von Baeyer, 2002) and the body has accommodated to this pain. Further research is needed.

In this study, parents commonly described various facial expressions as pain responses. This finding is consistent with several findings: (1) ICC for the facial subscale (0.81) of the Non-communicating Children’s Pain Checklist (Breau et al., 2002a); (2) the strong correlations in the Face and Cry categories in the FLACC (Voepel-Lewis et al., 2002); (3) the findings in a qualitative study in which parents identified facial expression as a strong indicator of pain (Carter et al., 2002).

The range of pain responses within the general categories of pain response was an important finding. For example, in response to pain, some children increased vocalizations while others decreased vocalizations. This has important clinical implications. Creating a pain assessment tool that captures the range of pain responses is challenging. The Non-communicating Children’s Pain Checklist includes extremes in pain response as separate items on the tool. Including each of the extremes of pain such as “body/limbs floppy” and “spastic tension rigid”; “less interaction, withdrawn” and “seeking comfort or physical closeness” “not moving, less active, quiet” and “jumping around agitated, fidgety” improves the comprehensiveness of this scale. However, if a child scores high in one item (for example, body/limbs floppy) then the child would score low in the opposite extreme (for example, spastic tension rigid). So when totaling the score for pain intensity, the items that depict extremes cancel each other out and do not contribute to the total pain score. Pain assessment tools that individualize, such as the Individual Numeric Rating Scale and the r-FLACC, allow for the range of pain responses observed in this population.

Another challenge of pain assessment in this population is that some of the descriptors for ‘no pain’ were responses to pain in other children. In this study 25% (n = 22) of the no pain descriptors (n = 88), such as “not vocalizing” or “active, moves a lot”, “seeks physical closeness”, “moans” or “sleeps”, described pain in other children. So a clinician, without knowing how a child responds when not having pain, may have difficulty differentiating a no pain state from a pain state. Both findings: (1) a range of responses to pain within general categories and; (2) that ‘no pain’ descriptors are pain descriptors in others support individualizing tools and documenting baseline behaviors for comparison during pain assessment.

It is a common assumption that pain expressions increase in number as pain intensity increases. Most pain assessment tools are based on this assumption. The findings from our study support the concept that the type of pain expression may change as pain intensity changes. For example, a child may express mild pain by grimacing but a change in muscle tone may indicate severe pain. Descriptors increased incrementally from mild to severe pain intensity only within one category: “changes in vocalization including crying”. In contrast, the following categories of pain responses peaked at moderate pain: physiologic, changes in muscle tone, and changes in social behaviors. Interestingly, the parents in this study reported more changes in facial expression with mild and moderate than with severe pain. It may be that children exhibit fewer facial descriptors when in severe pain. Another explanation is that the child’s escalating vocalizations during severe pain obscures the parent’s recall of facial changes.

Intellectual disability occurs as a result of many different physical conditions, such as prenatal events, infection, chromosomal abnormalities or even accidents such as near drowning or head injuries. Since the cause of intellectual disabilities often directly affects the child’s physical abilities and in some situations the source of pain, it follows logically that children with similar causes of intellectual disabilities would respond similarly to pain. In this study, there was a significant relationship between category of pain response and cause of intellectual disabilities (p < .001). Children whose cause of intellectual disabilities resulted from a prenatal or peri-natal event expressed pain using more facial expressions and fewer social and vocal descriptors than children with intellectual disability from other causes. Children whose cause of intellectual disabilities resulted from a seizure disorder had more physiologic and social and fewer facial descriptors. Further study with a large sample size and more varied causes of intellectual disabilities is needed to determine if the cause of intellectual disabilities predicts pain responses in children with similar causes of intellectual disabilities.

In this sample, there was a significant relationship between the sex of the child and categories of pain response $[\chi^2 (7, n = 423)=18.62, p = .01]$. Parents reported more physiologic and social responses in daughters and more changes in vocalization and fewer physiologic and social changes in sons. It is possible that the differences in pain expressions between males and females may be explained by sex differences. Many studies have found sex differences in pain response as well as responses to pain interventions. How to interpret these differences for nonverbal children with intellectual disability is difficult. The differences may be biological,
behavioral or both. Another explanation is that social and cultural influences may have confounded the data as it was collected from mostly mothers and by female researchers. Mothers and fathers may perceive their daughters pain responses differently than their son’s pain responses. Research is needed to further understand the impact of sex on pain responses in this population.

A challenge for all measurement tools is the balance between comprehensiveness and clinical utility. The Non-communicating Children’s Pain Checklist is the most comprehensive of the standardized tools. A limitation of the Non-communicating Children’s Pain Checklist is the current length of the 4 point likert checklist (30 items) which makes it burdensome for clinicians to complete every 4 h, which is the standard of care in most acute care settings. Although other tools had fewer items than the Non-communicating Children’s Pain Checklist, they did not account for, at best 17% (r-FLACC), at worst 32% (Pediatric Pain Profile and University of Wisconsin Children’s Hospital Pain Scale) of the parent described pain responses when comparing the items on the tools with the pain responses from this sample.

Individualizing pain assessment tools was introduced by Solodiuk and Curley (2003). Individualizing increases the comprehensiveness of a pain tool and allows for wide range of individual responses to pain. The 5 item structure of the r-FLACC, may limit its comprehensiveness only to the 5 categories of pain response (Face, Legs, Activity, Cry and Consolability). The 5 item structure limits the “weight” of the each of the individualized descriptors. For example, if a parent individualizes the third item in the r-FLACC, “activity”, to include the child’s response to severe pain, “he bites his right hand” this description would account for only 20% of the total score even though the parent identified this behavior as occurring in the presence of severe pain. In contrast, Individual Numeric Rating Scale allows documentation of baseline or no pain behavior; individualization without the structural limitations of standardized pain assessment tools and weighting behaviors that may differentiate severity of pain.

5. Limitations and clinical implications

There were several methodological limitations noted in this study. Common in a convenience sample, some patients and parents were underrepresented in the sample. For example, male parents, racial minorities and Non-English speaking parents were all underrepresented. As with all secondary analyses, the aims and research questions were limited by the data collected. Sufficient data was collected to examine the 3 aims of the study, but other questions had to be deferred. Research is needed to further validate and to measure the clinical utility of the Individual Numeric Rating Scale and to confirm this analysis.

This study expands on current knowledge in several ways. These results document the range of observable pain descriptors, some of which include extremes or opposite pain responses. It documents that no pain behaviors are pain descriptors in others. This study supports the Social Communication Model of Pain. Our findings corroborate the evidence that changes in vocalizations, social behaviors and facial expression are the most commonly identified pain descriptors in this population. This study provides preliminary evidence of the relationship between type of pain responses and severity of pain, cause of intellectual disabilities, and sex of child.

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References
