Parent Described Pain Cues in Nonverbal Children with Intellectual Disability: Deriving Patterns of Pain Responses and Potential Implications

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PARENT DESCRIBED PAIN CUES IN NONVERBAL CHILDREN WITH INTELLECTUAL DISABILITY:
DERIVING PATTERNS OF PAIN RESPONSES AND POTENTIAL IMPLICATIONS

a dissertation

by

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Abstract

Assessing pain in nonverbal children with intellectual disability (ID) is challenging. These children are at risk for having pain from complex medical conditions and treatments for these conditions (Breau, Camfield, McGrath, Finley, 2004). Compounding this, their pain cues are often misunderstood, given that they are nonverbal and limited by their physical abilities. Although, pain assessment tools for this population exist, there is a need for tools appropriate for a range of exhibited pain expressions.

The general purpose of this study was to examine the words that parents of children with ID use to describe their child’s pain responses in order to improve pain recognition and management. Specifically, the aims were to: 1) Identify common pain responses; 2) Examine the relationship between type of pain response and demographic characteristics; 3) Compare common pain responses to cues in the literature.

A non-directed summative content analysis identified patterns in 335 parent described pain responses of 50 nonverbal children with ID ages 6-18 years. The relationships between type of pain response and selected demographic factors were examined. Then pain responses were compared to items of pain tools for this population.

Seven distinct categories of pain expression were identified in the content analysis. The greatest percentage of pain cues were within the categories of vocalization (39.4%), social behavior (21.8%) and facial expressions (16%). Four categories: vocalization, social behavior, muscle tone and activity level included opposite responses to pain. Significant relationships between type of parent described pain expression and 1) pain severity; 2) causes of ID and; 3) the gender of the child found that type of pain expression changes with severity; that patients with seizure disorders expressed pain with
vocal pain expression; and that females expressed pain with more social pain expression while males expressed with more vocalizations.

The results support published evidence that parents can articulate their child’s pain responses. The study also provides evidence of: 1) opposite pain responses within general categories of pain; 2) a significant relationship between type of pain responses and severity of pain, cause of ID and child gender and; 3) the comprehensiveness of pain assessment tools vary greatly.
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There were several methodological limitations noted in this study. These limitations are: 1) the sole use of parent knowledge of the child for pain cues; 2) related to sampling of parents and children and; 2) related to the study being a secondary analysis. .................................................................................... 85

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Chapter 1: Statement of the Problem and Significance

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 (ID), assessment of behavior and other physical changes are the next best sources towards understanding their pain experiences. Accurate pain assessment of nonverbal children with ID is crucial because these children are at risk for having pain and for having pain that is misunderstood. Despite several published standardized pain assessment tools, at this time, none of these tools meets the requirements of a well established pain assessment tool as defined by Cohen, Greca, Blount, Kazak, Holmbeck, Lemanek, 2008. Also, there is still no consensus among clinicians caring for these children on the best way to assess pain in nonverbal children with ID. In addition to further testing of the published pain assessment tools, a better understanding of the nature of pain expression in this population is needed.

Purpose, Goal, Research Questions and Aims

This study drew upon parental knowledge of the child through an examination of the words that parents of a nonverbal child use to describe their child’s pain cues. The overall purpose of this study was to examine the words that parents of children with ID use to describe their child’s pain responses to identify patterns of pain responses that may be clinically useful during pain assessment. The goal of this study was to better understand the nature of pain expression in this population. The research questions were: What themes or patterns can be identified through an examination of parent reported descriptions of pain cues in nonverbal children with profound ID? What is the relationship between type of pain responses and the following independent variables:
child expressiveness, cause of ID, child gender, child developmental age and severity of pain and child co-morbidities and parent age? In relation to the purpose, goal and questions, the aims were to:

1) Identify common pain responses as described by parental report.

2) Examine the relationship between type of parent described pain responses of children with ID and the following independent variables: child expressiveness, cause of ID, child gender, child developmental age, severity of pain, child co-morbidities, and parent age.

3) Compare the parent identified pain responses to the pain cues described in the literature for this population.

This chapter provides an introduction to this study with definitions, assumptions and a description of the theoretical framework. Key definitions for this study were intellectual disabilities (ID), parent(s), the environment and patterns.

Definitions

Intellectual disability: a particular state of functioning that begins in childhood, in which limitations in intelligence coexist with limitations in adaptive skills (Schalock, Luckasson, Shogren, Borthwick-Duffy, Bradley, Buntinx, Coulter, et al., 2007). Throughout this study, the term “intellectual disability” is used to describe this population. This term, “intellectual disability”, is the preferred term for the disability formally referred to as mental retardation (Schalock, Luckasson, Shogren, Borthwick-Duffy, Bradely, Buntinx, Coulter, et al., 2007).

Parents: for the purpose of this study, were defined as the biologic, foster or adopted parents of children with ID.
Environment: was considered to be anything outside of the parent-child dyad.

Patterns: were defined as a reoccurring relationship between: 1) a pain descriptor and demographic information and/or the characteristics of the dyad or; 2) pain descriptors between and among patients.

Children with ID make up a small percentage of the general public, 0.3-2.5% of the United States population depending on how ID is defined (U.S Department of Education Statistics). However, children with ID are at risk for having health issues (van Schrojenstein Lantman-De Valk, Metsemakers, Haveman, Crebolder, 2000; Gilbert-MacLeod, Craig, Rocha, Mathias, 2000) and pain (Stallard, Williams, Lenton, Velleman, 2001; Hadden & von Baeyer, 2002). Therefore, pain recognition is critical to identifying the source of pain, treating health issues and providing safe, effective pain management.

Assumptions

This study was carried out with several assumptions. First, parents of children with ID have the knowledge to interpret their child’s pain cues. This parental knowledge of the child both when the child is in pain and when the child is in their usual state is advantageous to assessing pain accurately in this population. Parents become experts on their child’s pain cues with the hope of alleviating their child’s suffering. Without the benefit of verbal communication, changes in behaviors and expression are essential to understanding the experience of the nonverbal child.

Parental knowledge of the child develops over time. Repeated observation allows parents to develop insights related to their child’s pain, to intervene and watch the child’s responses to the interventions. An insight occurs when parents organize their knowledge by grouping together memories of similar responses and behaviors and coinciding
situations. For example, a child may suddenly stop eating. No other behavioral changes are noted. The parents try to feed the child his/her favorite food without success. The parents try other interventions such as changing the child’s position and putting on the child’s favorite music. Eventually, the parents bring the child to the pediatrician who discovers that the child has a bone fracture perhaps due to chronic steroid use for severe asthma. The bone is set and pain decreases. The child begins to eat. The parents remember that the child did not eat for days after the surgery. The parents have the insight that this child does not eat when in pain. Gradually, through long term observations, parents accumulate insights on how their child acts when in pain so that they can communicate to clinicians and advocate for their children.

These insights and observations eventually confirm or confuse, provoking further questions about pain assessment in their child. So the time parents spend with their child, in combination with the parental need to understand their child and to relieve suffering in their child contributes to parental insights and knowledge of their child’s pain cues.

The second assumption was that observed changes in behavior, facial expression and body posture do not occur randomly but each has a cause. There are many potential causes of behavioral changes in children with ID such as physiologic (for example as a result of electrolyte imbalances, or physical illness), a learned behavior that receives a certain response, or a way of communicating.

The third assumption was that the pain cues of nonverbal children with ID are diverse. Nonverbal children with ID are a heterogeneous population with a wide range of physical abilities and medical conditions. The wide range of physical abilities and medical conditions makes it difficult to identify a ‘usual’ response to pain in this
population. Researchers have identified common categories of responses but describe an array of individual responses within those categories (McGrath, Rosmus, Campbell, Hennigar, 1998; Terstegen, Koot, de Boer, Tibboel, 2003). Additionally, idiosyncratic responses to pain have been reported in this population (Fanurik, Koh, Schmitz, Conrad, 1999).

The fourth assumption was that pain in all human beings is important to treat using the best knowledge available. Effective treatment of pain improves the quality of life for the human being experiencing pain and his or her family. In the book, *Man’s Search for Meaning*, Frankl touches upon this topic. Frankl (1984) points out that society often judges the value of people based on their usefulness and achievement, while “value in the sense of dignity” is virtually ignored. This author writes from a similar philosophy, that treating suffering in all human beings acknowledges the “value in the sense of dignity” of all human beings.

**Theoretical Framework**

The theoretical framework for this study was adapted from Kathryn Barnard’s Child Health Assessment Model (Barnard, 1976). Although the Child Health Assessment Model was created for mother-infant interactions, it provides a framework for the interactions between parents and their nonverbal child with ID. The relationship of a parent and nonverbal child is similar to the relationship between mother-infant in that the child is not able to verbalize and the child is completely physically dependent on the parent. The relationship is different from the mother-infant interaction in that the parent-nonverbal child dyad has spent more time together, perhaps years, and therefore has had the time to learn how to better understand and respond to the other.
In the Child Health Assessment Model, Barnard depicted the interaction among the infant, the mother or caregivers, and the environment as three overlapping circles. For this study, the model was adapted to represent the child, the parent and the environment (See Figure 1). Within this model, the three parties have different responsibilities to maintain and foster the individuals and the relationships among the three. The nonverbal child with ID brings to the interaction and relationship: 1) physical abilities such as the ability to move body parts in a certain way; 2) expressive abilities (including clarity of cues and); 3) responsiveness to the parents or caregivers. The parent brings to the interaction: 1) the ability to comfort and alleviate distress; 2) receptive abilities (a seasoned sensitivity to the child’s pain cues); 3) the ability to interpret the child’s behaviors to others and 4) love for the child. Ideally, the environment would have resources to support the parent-child dyad. These resources may include knowledge of how to identify sources of pain and to manage that pain; resources that support time for the parents to care for themselves and their other responsibilities; and financial, social and emotional support.

Although the parent-child dyad is the main focus of the interaction as described by Barnard, the environment is also important. Barnard considered the environment to include extended family members, family friends, involved health care clinicians and society in general. For this population of children and families, health care clinicians are considered to be an important part of the environment because so many nonverbal children with ID have co-morbidities and see frequently see health care providers.
Figure 1

*Interpersonal Interaction: Informed Pain Assessment in Nonverbal Children with ID*
Pain Assessment and Management as Nursing Care

This research has significance for the nursing profession. Pain assessment and management is an important part of the “Central Unifying Focus” for the discipline of nursing: Facilitating Humanization, Meaning, Choice, Quality of Life, and Healing in Living and Dying” (Willis, Grace & Roy, 2008). Understanding a child’s pain experience through pain assessment is the first step in relieving the suffering caused by physical pain. In addition, understanding a child’s pain experience and relief of suffering facilitates humanization through recognition of the experience and efforts to manage suffering. Relief of suffering facilitates quality of life and healing.

Pain assessment and the subsequent management of pain is an important part of the role of nursing. In a hospital setting nurses complete and document most of the pain assessments for inpatients. Although other clinicians do assess pain (such as physicians, physical therapists, child life therapists), frequent nursing assessment of pain is required by hospital policy in most hospitals every four hours AND before and after interventions for pain in acute care facilities. Frequent nursing assessment of pain is required and monitored by hospital credentialing agencies such as The Joint Commission© (The Joint Commission, 2010). Nursing assessment and documentation of pain and interventions for pain is necessary to achieve and maintain Magnet Status through the Magnet Recognition Program®, a recognition program established by the American Nursing Credentialing Center (American Nursing Credentialing Center, 2010).

Although this study is an exploratory study, the results of this study will be helpful to clinicians to recognize the range of pain responses expressed by this population. It will helpful to clinicians to understand the relationships between pain
response and patient characteristics and to help to sort out the strengths and weakness of the published pain assessment tools for this population.
Chapter 2: Review of the Literature

This is a review of the literature for a study of parent described pain responses in nonverbal children with ID. This study drew upon parental knowledge of the child through an examination of the words that parents of a nonverbal child use to describe their child’s pain cues. The overall purpose of this study was to examine the words that parents of children with ID use to describe their child’s pain responses to identify patterns of pain responses that may be clinically useful during pain assessment. The goal of this study was to better understand the nature of pain expression in this population. The research questions were: What themes or patterns can be identified through an examination of parent reported descriptions of pain cues in nonverbal children with ID? What is the relationship between type of pain responses and the following independent variables: child expressiveness, cause of ID, child gender, child developmental age and severity of pain and child co-morbidities and parent age? In relation to the purpose, goal and questions, the aims were to:

1) Identify common pain responses as described by parental report.

2) Examine the relationship between type of parent described pain responses of children with ID and the following independent variables: child expressiveness, cause of ID, child gender, child developmental age, severity of pain, child co-morbidities, and parent age.

3) Compare the parent identified pain responses to the pain cues described in the literature for this population.

The review of the literature was structured in the following manner. First, general information about the incidence of pain in this population was reviewed. Then, parent
proxy pain assessment was discussed followed by a review of pain assessment tools tested specifically for this population. The tools are presented alphabetically and not chronologically since many of the studies were reported in the literature during the same 5-7 year period. Finally, the areas of emerging knowledge are stated with questions requiring further investigation.

**Scope of the Problem**

People with intellectual disabilities are more likely to have health problems than the general population. In a descriptive study comparing adults from the Netherlands with and without intellectual disabilities, logistic regression analysis found that the risk for health problems was 2.5 times greater in patients with intellectual disabilities (van Schrojenstein Lantman-De Valk, Metsemakers, Haveman, Crebolder, 2000). Similar results were found in children. In a study comparing the everyday pain responses in children with and without intellectual disabilities, children with intellectual disabilities had more medical experiences than children without intellectual disabilities (Gilbert-MacLeod, Craig, Rocha, Mathias, 2000). Both health problems and medical interventions were reported as sources of pain.

Unfortunately, pain in children with intellectual disabilities is common and rarely actively treated (Stallard, Williams, Lenton, Velleman, 2001). In a prospective study of nonverbal children with ID, 73.5% of the children (n=25) experienced pain on at least one day over a 2 week period as assessed by their parents and recorded in a diary. When the child had pain, the parent rated the intensity of the pain as mild, moderate or severe and the duration of the pain. Moderate to severe levels of pain were experienced daily by
68% of children with ID (Stallard, 2001). None of the children in the study was receiving treatment for their pain. The sources of the pain in this study were not documented.

Even when there is an obvious source of pain—such as surgical pain, patients with intellectual disabilities may not receive the interventions that others receive. In a study comparing the administration of opioids post operatively 1-3 days after a spinal fusion, children with intellectual disabilities received smaller total doses of opioids than those children without intellectual disabilities (Malviya, Voepel-Lewis, Tait, Merkel, Lauer, Munro, Farley, 2001). One possible reason for the under treatment of pain in this population is that the pain cues of these children, being nonverbal and limited by their physical abilities, may not be recognized or easily understood by others. Another reason may be that the amount of opioids estimated by clinicians to be safely administered may be reduced because of concerns of increased risk of opioid related respiratory depression due to the child’s co-morbidities and the central nervous system depressant medications given to treat these co-morbidities (for example, Baclofen and Diazepam given for spasticity).

Unfortunately, difficulties in pain recognition can have serious consequences. A study of deaths caused by intestinal obstructions found an unusually high mortality due to nonmalignant gastrointestinal obstruction in individuals with severe developmental disabilities (Roy & Simon, 1987; Jancar & Speller, 1994). Some have interpreted these tragic case studies as a sign of pain indifference or pain insensitivity (Biersdoff, 1994; 1991). However, other studies evaluating children with intellectual disabilities have found that although pain responses may be subtle or different than other people, pain cues in children with intellectual disabilities are identifiable (McGrath, 1998; Terstegen, Koot,
de Boer, Tibboel, 2003; Fanurik, Koh, Schmitz, Harrison, Conrad, 1999; Hunt, 2003; Carter, 2002; Defrin, Pick, Peretz, Carmeli, 2004). The increased mortality reported in the literature may be related to the lack of recognition of pain cues, leading to delays in medical evaluation in medically fragile people rather than pain insensitivity or indifference.

**Risk Factors for Pain**

Risk factors for pain in children with ID were described in a study of 94 children age 3-18 years with intellectual disabilities. Caregivers reported the children’s pain during weekly semi-structured telephone surveys for a one month period. Logistic regression analysis was then used to predict pain with the child’s characteristics and medical condition. During this study, caregivers described pain from: 1) accidents; 2) gastrointestinal issues; 3) musculoskeletal system; 4) common childhood sources such as teething, menstruation, and headache; 5) infections and 6) medical procedures such as from needle procedures, feeding tube irritation or surgery. In this population of 94 children with intellectual disabilities, 35% had pain weekly and on average, these children had 9-10 hours of pain per week (Breau, Camfield, McGrath, Finley, 2003a).

These authors further outlined risk factors for certain types of pain in children with ID (Breau, Camfield, McGrath, Finley, 2004). For example, not having visual or motor impairment increased the risk for accidental pain in this population. The authors explained the rationale for this as children without visual or motor impairment may be more independent and therefore more physically active and accident prone. Seizure disorder, leg impairment and greater number of medications increased the risk for non-
accidental pain. Being male and tube fed increased the risk of musculoskeletal pain.
(Breau, Camfield, McGrath, Finley, 2004).

**Parental Knowledge of Child’s Pain Responses**

Parental knowledge and expertise of the child has been used to assess pain in children with and without intellectual disabilities. Attempts at measuring parental expertise in the skill of pain assessment have shown mixed results. One study compared parent proxy pain assessment ratings and self reported pain assessments in healthy children (ages 7-12 years) undergoing day surgery procedures. Results showed a fairly close relationship between the parent and child score (n=110). However, kappa statistics indicated only poor to fair agreement beyond chance. The researchers concluded that for this population of healthy children, parental underestimation of pain may contribute to inadequate pain control (Chambers, Reid, Craig, McGrath, Finley, 1998). Similar results were demonstrated in a study comparing 63 emergency department patients (ages 4-7 years) with acute pain from trauma or the occurrence of procedural pain. There was poor agreement in pain ratings by healthy children’s self report, parents and practitioners (Singer, Gulla, Thode, 2002).

Mann, Jacobsen and Redd (1992) used regression analyses to further explore the perspectives of the children, parents and practitioners and the reasons they chose certain ratings. This study found that the nurse’s ratings were based on “overt distress” (p. 45). Parental ratings reflected parental knowledge of the child and deviation from usual behavior. The child’s self reported ratings of pain was associated with the child’s stage of development. The authors concluded that the differences in parent, child and nurse ratings seen in this study and in previous studies may be related to the differing
perspectives of the child, the parent and the nurse assessing pain rather than inaccuracies in measurement.

It may be that the results of the previously described studies that showed poor agreement in the parent and child pain scores do not reflect inaccuracies of parent proxy ratings for pain. The poor agreement may instead reflect the difficulties in matching scores based on a number scale that represents the spectrum of ratings between no pain and the “worst pain that you can imagine”. One individual’s worst pain imaginable may be very different from another’s based on past experiences, cultural background, and developmental level. An adult’s perception of worst pain imaginable is likely quite different from a healthy child’s version of worst pain imaginable. Although child perceptions and parent perceptions of pain are different, when self reports of pain are not possible, as is the case with nonverbal children with ID, then pain assessment by parent proxy is the next best source of pain assessment. Pain assessment by parent proxy is common in clinical practice. Pain assessment by parent proxy is described in *Pain Assessment in the Nonverbal Patient: Position Statement with Clinical Practice* (Herr, Coyne, Key, Manworren, McCaffery, Merkel, Pelosi-Kelly, Wild, 2006). This document recommends that credible information can be obtained from a parent or another person who know the patient well (Herr et al, 2006).

Parental expertise in pain assessment for verbal children capable of self report seems to be different than parental expertise for nonverbal children. Behavioral and physical pain cues are not as critical for parents of verbal children to recognize simply because the child can self report the pain experience. Parents of verbal children may have greater difficulty using behavioral pain cues because they primarily use the child’s
words for the basis of pain assessment. In nonverbal children with ID, parents only have behavioral and physical cues on which to base their pain assessment, so knowing these cues is critical.

Several studies of pain assessment in children with ID have found that parents can identify their child’s behavioral and physical pain cues (McGrath et al, 1998; Terstegen, Koot, de Boer, Tibboel, 2003; Fanurik, Koh, Schmitz, Harrison, Conrad, 1999; Hunt, Mastroyannopoulou, Goldman, Seers, 2003; Carter, McArthur, Cunliffe, 2002). Voepel-Lewis et al (2005) studied the validity of parent pain ratings in children with cognitive impairment. Parent’s scores correlated well with nurse ratings: intraclass correlation coefficient=0.78 [confidence interval =0.63-0.87] and intraclass correlation coefficient =0.73 [confidence interval 0.59-0.83] respectively.

Carter et al (2002) described the process of how parents become experts of their child’s pain cues over time in a qualitative case study design with 15 parents of children with ID. Parents described 3 processes used in assessing pain: “guessing about the pain, working it out and instinctively or intuitively knowing” (p. 452). “Guessing” about the pain would be used in the early months when parents were learning their child’s pain cues. Guessing was 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 became expert of their child’s pain and then internalized the process so that they could identify pain cues and possible causes quickly. In this study, all the parents based their pain assessments on a change from the child’s usual behavior.
Hunt et al (2003) used a grounded theory approach to explore how parents and practitioners assessed pain in nonverbal children with ID. Parents of nonverbal children with ID (n=21) and practitioners who care for this population (n=26) were interviewed regarding the child’s history of pain, pain behaviors, processes by which participants assessed pain and provided or sought treatment for the pain. Three forms of knowledge emerged as critical to pain assessment in this population: 1) knowledge of the child; 2) familiarity with children with similar conditions and; 3) knowing the science (Hunt et al, 2003).

Another study reviewed the beliefs about pain that parents and caregivers of children with cognitive impairment have. Parents (n=52) and caregivers (n=13) completed the Mental Retardation Attitude Inventory- Revised and the Pain Opinion Questionnaire. The Mental Retardation Attitude Inventory- Revised assesses attitudes towards people with intellectual disabilities using a 6 point likert scale. The Pain Opinion Questionnaire, designed for this study, addresses 5 facets of pain: sensation, emotional reaction, behavioral reaction, communication and frequency. The results of this study found that parents and caregivers believed “children’s pain sensation becomes greater, relative to children without ID, as severity of ID increases and that pain reaction is most consistent with pain sensation for children with severe ID” (Breau, 2003c, p. 343). Further research is needed to replicate these findings and to understand what these beliefs are based upon.
Pain Responses

Several teams of researchers have begun to describe how children with intellectual disabilities respond and react to pain. Responses to pain in this population have been described as: 1) individual (McGrath, 1998; Terstegen, Koot, de Boer, Tibboel, 2003; Carter, 2002); 2) idiosyncratic (Fanurik et al, 1999); and 3) dampened and delayed (Orberlander, Gilbert, Chambers, O’Donnell, Craig, 1999). Each one of these responses reported in the literature will be described in greater detail.

McGrath (1998) reported that individual responses to pain were common in this population. These responses could be grouped into seven general categories including: 1) Vocal; 2) Eating/Sleeping; 3) Social/Personality; 4) Facial expression of pain; 5) Activity; 6) Body and Limbs; 7) Physiological. In the same study, he reported that “because of the individual responses to pain, there may not be a single set of items that can be reliably used to discriminate pain in this population” (McGrath, 1998, p. 342).

Similarly, Terstegen (2003) described the individual responses to pain in this population. Pain indicators of 52 children with surgical pain and ID were identified through interviews with parents, and clinicians and through observations of the children during observed painful events (dental procedures, physical therapy and vaccinations) (n=25). Of the 138 indicators identified, 30 were sensitive to pain using levels of significance measured from 2 researchers’ assessments in post surgical video tapes of 52 children. They concluded: “although specific expression of pain may be highly individual, there appears to be a set of shared generic indicators” (Terstegen, 2003, p. 197). The shared generic indicators are grouped under the following headings: (1) facial expression; (2) motor behavior; (3) social behavior/ mood; (4) attitude toward sore body
part; (5) vocalization; (6) physiological signs (Terstegen, 2003). Carter et al (2002) reported similar results as parents described a diversity of pain responses in their children with some commonalities or similarities identified among the children.

Another study, undertook semi-structured interviews with 29 female caregivers (25 of which were the mother) of non communicating children. The caregivers identified 203 separate pain cues for the children they cared for. A 2 stage Delphi process was undertaken with a group of experts (3 clinical psychologists, 1 pediatrician, 1 nurse, 2 researchers, 2 parents of non communicating children) to categorize the 203 pain cues into 11 groups (Stallard, Williams, Velleman, Lenton, McGrath, 2002). The 11 categories were then compared to what caregivers identified as 95 cues indicating definite pain and 68 cues indicating severe pain. One or more of 6 cues (“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”) were identified by 90% of caregivers as signs of pain (Stallard et al, 2002, p. 146). However, given how wide ranging some of the categories are, such as “screaming, yelling, groaning or moaning” or “screwed up or distressed looking face”, this result is not unexpected. For example, “screwed up or distressed looking face” could be interpreted in many different ways such as by changes in the eyes, the nose, the mouth, or the entire facial expression to a panel of experts.

A second study by this team found that 20 percent of parents or caregivers described pain responses in their child that were not described in the 6 items of the Pain Indicator for Communicatively Impaired Children (PICIC) (Stallard, Williams, Velleman, Lenton, McGrath, Taylor, 2002a). In addition, the study reported a significant difference
in the usage of the core items and the parent identified pain cues ($X^2=26.67$, df=2, $p<0.001$). The parent identified pain cues were used more frequently to assess pain (35.4% of observations). The 6 core items of the PICIC pain assessment tools were used in 22.9% of the observations. Further research on this tool is needed to test whether clinicians with little previous knowledge of the child, such as nurses in an acute care setting, would elicit the same results.

Fanurik (1999) described pain responses as sometimes idiosyncratic in this population. In interviews of parents of children with ID, Fanurik (1999) reported that sometimes the pain behaviors described by the parents were unusual or even idiosyncratic such as “makes an ‘O’ with her mouth and sticks out her tongue”; “gets quiet and still”, “he growls”; “red spots on his face”, “bites his left hand when hurting” (Fanurik et al, 1999, p. 231).

Self-injurious behavior is a common form of idiosyncratic behavior sometimes exhibited in this population of nonverbal children with ID. It is estimated that 2%-50% of people of all ages with this disability have self-injurious behavior. In an exploratory study of self-injurious behavior in this population, multivariate analysis of variance indicated that pain scores did not differ between children with and those without self-injurious behavior. These findings do not support the hypothesis that children with self-injurious behaviors have a reduced response to pain (Breau, Camfield, Symons, Bodfish, Mackay, Finley, McGrath, (2003).

Oberlander (1999) reported that behavioral and physiologic pain responses as measured by heart rate and videotaped facial action assessments were dampened and delayed in adolescents with intellectual disability during routine vaccinations. In this
study, there were only modest changes in the VAS scores assessed by third-party blinded coders (Oberlander, 1999). Further study is needed to determine if parents or caregivers would be able to detect the child’s pain responses during immunizations.

**Pain Assessment Tools**

There has been increasing research on creating pain assessment tools for this population. Currently there are 6 tools published by teams of researchers in the English Language for this population (See Table 1). The Echelle Douleur Enfant San Salvador (DESS) was not included in the descriptions because the English version of the tool has not been validated. None of these 7 tools, including the DESS, can be described as a well established assessment tool as defined by Cohen, Greca, Blount, Kazak, Holmbeck, Lemanek, 2008. A well established assessment tool is a tool that has been: 1) “presented in at least 2 peer-reviewed articles by different investigators or investigatory teams; 2) [presented with] sufficient detail to allow for critical evaluation and replication and 3) [presented with] detailed information about the reliability and validity in at least one peer reviewed journal” (Cohen, 2008, p. 913).

Although the Noncommunicating Children’s Pain Checklist (NCCPC) has a greater amount of peer reviewed articles, the NCCPC in addition to the other tools described below have not been tested by different researchers or investigatory teams. The following text outlines each of the 6 published pain assessment tools tested in this population.

**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, Voepel-Lewis, Shayevitz, Malviya,
29

1997). This tool has been studied for children with intellectual disabilities (Malviya, Voepel-Lewis, Burke, Merkel, Tait, 2005; Voepel-Lewis, Merkel, Tait, Trzcinka, Malviya, 2002; Merkel, Voelpel-Lewis, Shayevitz, Malviya, 1997). In the first study of the FLACC for children with intellectual ability, the tool was not modified (Voepel-Lewis 2002). The FLACC scores by nurses correlated with parent scores (p< 0.001). The FLACC scores decreased after administration of analgesics (p< 0.001). In this study, the best correlation in scores were in categories of Face and Cry (r=0.505-0.698, k=0.3.3-0.448 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). Given the population of children with varying degrees of intellectual and physical abilities, the low correlation in the legs category is likely related to limited physical mobility in this population.

Although the study showed good validity and reliability, the sample chosen for this study had a wide range of ID. 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. This population of children with a wide variety of verbal abilities and intellectual disabilities may have skewed the results by including a large percentage of children with a high level of expressive abilities. Perhaps 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 intellectual disabilities. Further study is needed to determine if the same results would be found in a sample of nonverbal children with ID.

The FLACC was revised to include parent identified pain cues. Initial findings support the reliability and validity of the revised FLACC as a measure of pain in children
with cognitive impairment (intraclass correlation coefficient = 0.87-0.92; criterion validity r=0.78-0.87; construct validity using paired t tests decrease in pre to post analgesic pain scores (p<0.001) (Malviya et al., 2005). However, again, the study sample had a wide variety of ID [only 46% (n=24) with significant ID; 23% (n=12) had moderate impairment and 31% (n=16) had mild impairment. Twelve of these children were able to self report. So it is unknown whether the positive results were influenced by assessing children with a high level of expressive and physical abilities.

The 5 item structure of the FLACC limits the weight that parental knowledge has on the final pain score. For example, a parent individualizes the third item in the FLACC, “activity”, to include the child’s response to severe pain, “he bites his right hand”. Using the FLACC, 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.

The clinical utility of the FLACC was studied by the original authors. Nurses (n=15) and physicians (n=5) reviewed video tapes of children in pain and then rated the clinical utility of the FLACC as high in clinical utility as compared to the Non-Communicating Children’s Pain Checklist (NCCPC) (Voepel-lewis, Malviya, Tait, Merkel, Foster, Krane, 2008). This study would be strengthened by including the clinical condition and level of ID of the children in the reviewed videotapes, since this information would impact how clinicians rated the clinical utility of the tools. Also, reporting the demographics of the clinicians [nurses (n=15) and physicians (n=5) from 3 different sites and a variety of specialties] and how they were chosen to evaluate the tools for clinical utility would strengthen this study. Despite these limitations of the study, the
familiarly that most pediatric nurses have with the FLACC does increase its clinical utility.

**Individualized Numeric Rating Scale (INRS).**

The INRS is a horizontal numeric rating scale with open space for parents to populate the child’s usual behavior when not in pain and the child’s pain cues. Solodiuk and Curley (2003) introduced the concept of individualizing pain tools for nonverbal children with ID in response to the wide range of pain responses and physical abilities described in the literature and observed in clinical practice.

Initial testing of the INRS found that all the parents of 50 nonverbal children with severe ID were able to complete the task of describing and rank ordering their child’s behavior when in usual state and when having pain. Inter-rater reliability of the resultant INRS was tested across triads of raters (parent, bedside nurse and research nurse) in a total of 170 assessments completed 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-.87) across all ratings. Parent and bedside nurse agreement for ratings immediately before a pain intervention (ICC=.65) and post intervention (ICC=0.74) also suggested good reliability. Agreement between bedside nurse and research nurse (ICC 0.74-80) also suggest good reliability. The study also reported moderate to strong correlation between INRS and Non-communicating Children’s Pain Checklist- Post Operative Version (0.63-0.73) (Solodiuk, Scott-Sutherland, Meyers, Myette, Shusterman, Karian, Harris, Curley, 2010). These results provide preliminary data that the INRS is a valid and reliable tool for assessing pain in this population.
Non-communicating Children’s Pain Checklist (NCCPC).

The most studied tool for this population is the NCCPC (McGrath, Rosmus, Campbell, Hennigar, 1998; Breau, Finley, McGrath, Camfield, 2002; Breau, McGrath, Camfield, Finley, 2002a; Breau, Camfield, McGrath, Rosmus, Finley, 2001; Breau, McGrath, Camfield, Rosmus, Finley, 2000). This 31 item checklist was originally created from the descriptions of pain cues reported in interviews of 20 parents or caregivers of children with cognitive impairments (McGrath, 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 nonpainful but distressing situation and; 4) when the child was calm (Breau, McGrath, Camfield, Rosmus, Finley, 2000). More pain cues were present during pain when compared to calm situation; however the cues did not differentiate between pain and being in distress but not in pain.

The later version of this checklist, the NCCPC-PV (Non-communicating Children’s Pain Checklist – Postoperative Version) contains 27-items classified into six subscales (Breau, Finley, McGrath, Camfield, 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). The tool showed good interrater reliability in some of the categories post operatively (intra-class correlation coefficients 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 moderate correlations in most of the caregiver and researcher scores. One exception was the correlation of the caregivers
and researchers scores of the before surgery, which were not significant. Also, there were no significant correlations between the visual analog scale of nurses and the NCCPC-PV 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 the use of the tool by nurses.

Perhaps in an effort to include a wide range of individual responses to pain, some of the items describe opposite responses to pain. For example, some of these items describing opposite responses to pain are: “less interaction, withdrawn” versus “seeks comfort and physical closeness”; “not moving, less active, quiet” versus “jumping around, agitated, fidgety”; “body/limbs floppy” versus “stiff spastic, tense or rigid” (McGrath, 1998, p. 342). When evaluating these responses, the extremes may cancel each other out and therefore decrease the importance of each item in the total score. For example, if a child is observed as having body/limbs floppy very often (item score of 3) then it is likely that the child will be scored as having a stiff, spastic tense or rigid body not at all (item score of 0). Another limitation of this tool is the current length of the 4 point likert checklist (27 items) makes it burdensome for clinicians to complete every 4 hours, which is the standard of care for patients at risk for pain in most acute care settings. Despite these limitations, this tool is an important step towards developing a pain tool for this population.

**Pain Indicator for Communicatively Impaired Children (PICIC).**

The PICIC is a pain tool created for this population of children (Stallard, et al, 2002). The one study testing the psychometrics of this tool identified 6 pain cues for this population. These are “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”.

In this study, parents assessed pain at 14 different times over a one week period of time. 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). This finding supports previous reported evidence of the importance of facial expression of pain in this population. It is consistent with the following reports: 1) intra-class correlation coefficients for the facial subscale (0.81) of the NCCPC (Breau et al., 2002b); 2) the strong correlations in the Face and Cry categories in the FLACC (Voepel-Lewis et al., 2002); 3) the literature on the primal face of pain (Schiavenato, 2008) and 4) the findings in a qualitative study in which parents identified facial expression as a strong indicator of pain (Carter et al. 2002).

Several questions remain, such as since “screwed up or distressed looking face” describes more of a change in facial expression than a description of a specific response, exactly what did the parents base their assessment on? Was it the eyes or the eyebrows or the mouth or other facial expressions? Or was it a combination of these? In addition, little demographic information was reported about the 49 subject sample except for age, sex and general categories of etiology of ID. Additional information that would be helpful includes a measurement of the developmental level of the child and the child’s expressive abilities. Further research is needed to determine whether this information is generalizable.
Pediatric Pain Profile (PPP).

The Pediatric Pain Profile is a 20-item pain assessment tool. It was developed based on parent interviews of their child’s pain cues for children with ID. Qualitative interviews with parents and caregivers guided the development of this behavioral standardized pain assessment tool. Initial psychometric testing was completed using video filmed observations of the children in their usual setting during their everyday morning activities. Pain assessments were compared with saliva cortisol levels. The author concluded that cortisol levels are not a useful criterion for pain in this population (Hunt, 2007).

Interrater reliability testing of the PPP 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). Another study compared the pain assessments of the investigators with the pain assessments of experienced nurses caring for children with intellectual disabilities observed via videotapes and the nurses were allowed to replay the videos if needed. Since these study environments do not mimic the acute care clinical environment, it is difficult to determine whether these results are generalizable to a clinical environment. Also, further testing is needed to determine whether the results would be the same if clinicians without prior knowledge of the child or familiarity with the population were assessing the patient.

University of Wisconsin Children’s Hospital Pain Scale (UWCH).

The University of Wisconsin Children’s Hospital Pain Scale is a pain assessment tool created for preverbal and nonverbal children (Soetenga, Frank, Pellino, 1999). Initial testing of the scale showed good validity (internal consistency =0.93) and reliability
(interrater reliability =0.92). However, the small number of nonverbal children with ID (n=15) which was combined with preverbal children (n=59) makes it difficult to evaluate its usefulness for children with intellectual disabilities.
Table 1  *Comparison of Pain Assessment Tools for Nonverbal Children with ID*

<table>
<thead>
<tr>
<th>Tool</th>
<th>Type</th>
<th>Items</th>
<th>Reliability</th>
<th>Validity</th>
<th>Strength</th>
<th>Weakness</th>
</tr>
</thead>
<tbody>
<tr>
<td>FLACC r-FLACC</td>
<td>Behavior</td>
<td>5</td>
<td>ICC= 0.87-0.92</td>
<td>Construct validity p=0.001. Criterion validity r= 0.78-0.87.</td>
<td>Ease of use. Familiar.</td>
<td>Range of ID in sample may skew results. No baseline. Limited testing for nurses.</td>
</tr>
<tr>
<td>INRS</td>
<td>Child specific Responses</td>
<td>Child Specific</td>
<td>IRR parent/RA 0.74 Parent/RN ICC =0.65</td>
<td>Convergent validity with NCCPC-PV r=.63-.73.</td>
<td>Weighted behaviors. Ease of use. Includes baseline.</td>
<td>Time to populate.</td>
</tr>
<tr>
<td>NCCPC-R NCCPC-PV</td>
<td>Behavior Physical</td>
<td>R 30 PV 27</td>
<td>ICC= 0.78-0.82</td>
<td>Construct validity p&lt;0.01. Criterion validity r= 0.39-0.72.</td>
<td>Well studied.</td>
<td>Lengthy. No baseline. Needs further testing for nursing use.</td>
</tr>
</tbody>
</table>
Table 1 *Comparison of Pain Assessment Tools for Nonverbal Children with ID*  
*(Continued)*

<table>
<thead>
<tr>
<th><strong>PICIC</strong></th>
<th>Behavior</th>
<th>Sleep</th>
<th>Seizures</th>
<th>6</th>
<th>Not measured.</th>
<th>Not measured</th>
<th>Ease of Use.</th>
<th>Needs further study. No baseline. Needs further testing for nursing use.</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>PPP</strong></td>
<td>Behavior</td>
<td>Sleep</td>
<td>Seizures</td>
<td>20</td>
<td>ICC=0.74-0.89</td>
<td>Construct validity p&lt;0.001</td>
<td>Ease of use. Includes baseline.</td>
<td>Reversed scoring. Lengthy. Needs further testing for nursing use.</td>
</tr>
<tr>
<td><strong>UWCH</strong></td>
<td>Behavior</td>
<td>Sleep</td>
<td></td>
<td>5</td>
<td>IRR=0.92</td>
<td>Internal Consistency 0.93</td>
<td>Ease of use.</td>
<td>Sample may have skewed results. Needs further study. Global rating. No baseline.</td>
</tr>
</tbody>
</table>

**ICC** Intraclass Correlation  
**IRR** Interrater Reliability
Summary of Literature

Pain assessment in nonverbal children with ID is challenging because the child is at risk for having pain and cannot verbalize the pain experience. In the last decade, pain responses in this population have been studied by several research teams. From these studies, evidence is mounting in the following areas.

1. Parents of children with intellectual disabilities provide reasonable estimates of their child’s pain (Voepel-Lewis, Malviya, Tait, 2005). Parents are able to articulate pain behaviors in their children with intellectual disabilities (Solodiuk, 2010, Solodiuk, 2003; Fanurik, Koh, Schmitz, Harrison, Conrad, 1999;). In fact, most pain tools for this population were created from parent identified pain cues (McGrath, 1998; Terstegen, Koot, de Boer, Tibboel, 2003; Hunt, 2003; Carter, 2002). Using parental knowledge of a child’s pain cues is common in clinical practice. It is consistent with the statement in Pain Assessment in the Nonverbal Patients: Position Statement with Clinical Practice, which recommends that credible information, can be obtained from a parent or another person who knows the patient well (Herr, Coyne, Key, Manworren, McCaffrey, Merkel, Pelosi-Kelly, Wild, 2006).

2. Personal knowledge of a child is helpful in order to compare a possible response to pain with past pain behavior and a deviation from the usual behavior patterns (Carter, 2002; Hunt, 2003).

3. Changes in facial expression are critical to pain assessment in this population (Stallard et al, 2002; Breau et al, 2002; Voepel-Lewis, 2002; Carter, 2002; Schiavenato, 2008).
Although several groups of researchers have published data on the psychometric properties of pain tools in this population, at this time, none of the standardized pain assessment tools meets the requirements of a well established pain assessment tool as defined by Cohen, Greca, Blount, Kazak, Holmbeck, Lemanek, 2008. In addition, further testing of pain assessment tools for nonverbal children with intellectual disabilities should include a sample population with a large number of nonverbal children with ID. Some of the pain assessment tools described in the literature were tested in a sample population with a wide range of ID. Some even tested children with mild ID who could communicate their pain verbally. This type of sampling likely skewed the results of psychometric testing.

Another limitation of many of the published studies is that interrater reliability was measured comparing pain assessment scores of parents and researchers, but not including the scores of bedside nurses. There is mounting evidence that parents of children with ID can assess pain accurately in their child using their prior knowledge of the child. Researchers with their knowledge of the pain assessment in this population are likely to have more knowledge than most clinicians in assessing pain in this population. So it is not surprising that the psychometric testing showed positive results. An important clinical requirement is finding a pain assessment tool that bedside nurses can use to assess pain. So further testing of these tools with bedside nurses is important. Several of the pain assessment tools published in the literature are quite long so the clinical utility of these tools is limited for use in an acute care clinical setting in which assessing pain every 4 hours, before and after interventions for pain, is the standard of care.
Despite the published research, described in this chapter, the following general questions remain:

1. Which tools can be used by nurses in a hospital setting? As noted, most of the tools for this population were tested by parents or researchers. These tools may be helpful for parents to use at home. However, studies have not shown that parental assessment using behavioral pain tools is any more accurate than a parental estimate of pain without a pain assessment tool. Parents are not always able to stay at a hospital to assess pain. In most hospital settings, nurses have the responsibility to assess and document pain assessment regularly. The gap in the literature is pain tools tested in a hospital setting by nurses using perceived parental cues.

2. How to stimulate parental insights of their child’s pain cues and to document these observations in a manner that is accessible and useable to others? As noted, this is important since prior knowledge of the child is helpful to assessing pain.

3. If further cases of pain insensitivity or indifference are identified, what methods can be used to study these patients to determine whether pain is sensed but responses are obscured or to find ways to keep these patients safe if pain is indeed not sensed?

4. What patterns of pain responses can be detected through an examination of parent described pain responses in this population?

This last question is the primary question of this exploratory study. This study will add to this body of knowledge in the following ways. First, examining the words that parent’s use to describe their child’s pain will help us to examine common pain
responses, how often these responses are seen in this population and if specific categories of children exhibit certain responses to pain. Second, comparing the pain cues found in published pain assessment tools with the collected data from this study will serve to integrate the current knowledge and propose approaches to best practice.
Chapter 3: Methods

This is an explanation of the methods used for a study of parent described pain responses in nonverbal children with ID. This study drew upon parental knowledge of the child through an examination of the words that parents of nonverbal children use to describe their child’s pain cues. The overall purpose of this study was to examine the words that parents of children with ID use to describe their child’s pain responses to identify patterns of pain responses that may be clinically useful during pain assessment. The goal of this study was to better understand the nature of pain expression in this population. The research questions were: What themes or patterns can be identified through an examination of parent reported descriptions of pain cues in nonverbal children with ID? What is the relationship between type of pain responses and the following independent variables: child expressiveness, cause of ID, child gender, child developmental age and severity of pain and child co-morbidities and parent age? In relation to the purpose, goal and questions, the aims were to:

1) Identify common pain responses as described by parental report.

2) Examine the relationship between type of parent described pain responses of children with ID and the following independent variables: child expressiveness, cause of ID, child gender, child developmental age, severity of pain, child co-morbidities and parent age.

3) Compare the parent identified pain responses to the pain cues described in the literature for this population.
Introduction to the method: Summative qualitative content analysis

As described by Hsieh & Shannon (2005), a summative qualitative content analysis includes counts of words or phrases and also a content analysis. A content analysis is the process of interpreting, comparing and classifying the content into themes and patterns (Hsieh & Shannon, 2005). This technique has been used to evaluate a variety of topics such as the content of nursing textbooks related to death and dying (Ferrell, Virani, Grant, Borneman, 1999) or the content of lumbar spine x-ray reports (Thompson, Carr, 2007). This method has not been recorded in the literature as method to analyze parent descriptors of pain responses. This method is appropriate to study the previously listed study aims.

A summative qualitative content analysis of parent identified descriptors and the descriptors in published tools will add to this body of knowledge about pain assessment of nonverbal children with ID in the following ways. First, examining the words that parent’s use to describe their child’s pain will help to identify common pain responses and whether or not children with certain characteristics exhibit certain responses to pain. Secondly, comparing the extensive list of pain cues described by parents with the pain descriptors in published pain assessment tools will serve to evaluate the comprehensiveness of the pain tools.

Setting

This study is a secondary analysis of data obtained during research evaluating the validity and reliability testing of the INRS (Individualized Numeric Rating Scale) (Solodiuk, 2010). The setting for this study was in a 350 bed acute care pediatric hospital in the Northeast. Specifically, the discussions with parents about their child’s pain
responses were completed either in an exam room in the Preoperative Clinic several days before the child’s elective surgery or in the child’s room in one of the surgical inpatient units after surgery.

Sample

With approval from the hospital’s institutional review board (IRB) and parental consent, data was collected for the original study on a prospective, convenience sampling of 50 parents of nonverbal children with ID (ages 6-18). The inclusion criteria were: 1) parent had a child age 6-18 years with ID; 2) the above described child was scheduled for elective surgery requiring an overnight stay in the hospital and; 3) the parent was planning to stay at the hospital with the child. Exclusion criteria were: 1) the above-described children could communicate verbally even with one word or via a communication board and; 2) children were not currently living with their families, or had not lived with their families in the last 6 months. As part of the informed consent, parents were reminded that they could withdraw at any time without affecting their child’s care.

Potential subjects were screened for the above inclusion and exclusion criteria and informed of the study by a preoperative clinic nurse. If the parents were interested, the principal investigator (PI) or a research assistant (RA) would discuss the study in detail and offer consent. Once enrolled, the PI collected demographic data by using existing data in the medical record and/or by parent interview as shown in Table 2.
Table 2

Demographics Collected

<table>
<thead>
<tr>
<th>Child Demographics</th>
<th>Parent Demographics</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age</td>
<td>Age</td>
</tr>
<tr>
<td>Gender</td>
<td>Gender</td>
</tr>
<tr>
<td>Race and Ethnicity</td>
<td>Race and Ethnicity</td>
</tr>
<tr>
<td>Developmental Level</td>
<td>Educational Level</td>
</tr>
<tr>
<td>Cause of ID</td>
<td>Income</td>
</tr>
<tr>
<td>Co-Morbidities</td>
<td></td>
</tr>
<tr>
<td>Reason for surgery</td>
<td></td>
</tr>
<tr>
<td>Type of surgery</td>
<td></td>
</tr>
<tr>
<td>Date of surgery</td>
<td></td>
</tr>
</tbody>
</table>

Vineland Adaptive Behavior Scale.

In the original research data collection, the child’s level of functioning was measured by the Vineland Adaptive Behavior Scale (VABS). The administration of the VABS was completed through a semi structured interview of the parent by a specially trained psychologist. The VABS measures the child’s level of functioning in the following 4 domains: communication, daily living skills, socialization and motor skills. The scale consists of 301 items listed developmentally. A domain is considered complete when 7 consecutive items are scored as 0. The psychometrics of the VABS are internal consistency (split-half 0.76-0.99), with test retest reliability of 0.76-0.93, and interrater reliability of 0.62-0.78 (Rosebaum, 1995; Vig, Jedrysek, 1995) as shown in Table 3.
Data from the VABS (the total score and the communication domain subscale) were used for the chi square analysis.

Table 3

_ Vineland Adaptive Behavior Scale_

<table>
<thead>
<tr>
<th>Administration</th>
<th>Semi structured interview</th>
</tr>
</thead>
<tbody>
<tr>
<td>Type of response</td>
<td>Likert responses</td>
</tr>
<tr>
<td>4 domains</td>
<td>Communication</td>
</tr>
<tr>
<td></td>
<td>Daily Living Skills</td>
</tr>
<tr>
<td></td>
<td>Socialization</td>
</tr>
<tr>
<td></td>
<td>Motor Skills</td>
</tr>
<tr>
<td>Total items</td>
<td>301 items listed developmentally</td>
</tr>
<tr>
<td></td>
<td>if 7 consecutive items scored 0, skip to next domain</td>
</tr>
<tr>
<td>Reliability</td>
<td>Internal consistency (split-half) 0.76 to 0.99</td>
</tr>
<tr>
<td>(Rosenbaum, 1995; Vig &amp; Jedrysek, 1995)</td>
<td>Test-retest reliability 0.76 to 0.93</td>
</tr>
<tr>
<td></td>
<td>Interrater reliability 0.62-0.78</td>
</tr>
<tr>
<td>Validity</td>
<td>Correlation between Vineland II and VABS 0.65-0.94</td>
</tr>
<tr>
<td>(Sparrow et al, 2005)</td>
<td></td>
</tr>
</tbody>
</table>

**Sample Size.**

The number of participants involved in the original research study (n=50) was determined by conducting a power analysis. Although this sample size is large for a content analysis in a qualitative study, it seems adequate for the purposes of this study for the following reasons. First, upon review of the literature of pain tools for this
population, an n=50 would be the largest sample size of children who were nonverbal with ID. Currently, the largest number of nonverbal children with ID studied for pain assessment techniques is 40 (from a total sample of n=79 or 51% of the total sample). This before mentioned sample also included children who had “good verbal communication” and “minimal communication with simple words or signs” (Voepel-Lewis, et al 2002, p. 1227).

Second, since one of the aims of the study is to analyze relationships between variables, the larger number is required for the statistical analysis. Larger numbers are especially needed for the independent variables that have smaller numbers in certain categories.

Third, in clinical observations, this number seems adequate, because although children sometimes exhibit unique responses to pain, the number of possible responses is not infinite. Responses are limited by the body (the number of accessible body parts to respond with) and the child’s physical abilities. For example, self injurious pain expressions are limited by the method of injury, such as pulling skin or hair, scratching or biting or striking body parts against a hard object, and the number of body parts accessible to injury, such as upper and lower extremities, head, hair and accessible skin.

The proposal for this secondary analysis was reviewed by and approved by the hospital’s IRB and the university’s IRB. Throughout the planning and data collection phases of the original study, the ethics of investigating a vulnerable population were considered. To ensure that the consent process was consistent and thorough, only the PI and 2 RAs offered consent to the parents. Efforts were made throughout the data collection process to develop a respectful and cooperative research relationship that
would encourage disclosure, trust and awareness of potential ethics issues (Orb, Eisenhauer, Wynaden, 2001). Parents sometimes revealed unresolved issues related to the child’s complex postoperative medical care. When this occurred, the PI contacted and discussed the issue with the appropriate clinicians, with parental agreement.

**Data Collection Plan**

In the original study, semi-structured face-to-face interviews were conducted in a private room within the hospital by the preoperative clinic nurse or the PI, after obtaining informed consent from each parent. Guided by the aims of the study, only the parent’s words describing the child’s pain responses were transcribed onto the INRS form at the time of the interview.

The parent was asked to first describe their child’s behavior when not in pain on a usual day. Then the parent was asked to recall a time when they knew their child had pain whether from past surgeries, painful procedures and/or experiences. Parents were asked to describe those responses and link them with a number from 1-10 with 10 indicating severe pain. To help parents recall past pain responses, nurses used the FLACC (Face, Legs, Activity, Cry and Consolability) acronym. For instance, nurses asked the parents to think about how the child expressed pain with the face, legs, with changes in activity level, with changes in vocalizations and by how easily the child is consoled. This question was followed by probing questions (such as: “How did the child’s expression change?”; “How did the child move his/her arms?”) were asked to further explore the parental responses. The parental responses were then used to populate the INRS, a pain tool created specifically for the child from the parents’ descriptors or prior pain responses.
After the form was populated, the parents were asked to review the populated INRS and to make any changes to the tool. During subsequent post operative meetings with the parents to collect data for the primary study, the parents were once again asked to review the populated INRS and either to confirm or make any changes to the tool in order to verify and if necessary, clarify the collected data.

Data Analysis

Data analysis is a process of interpreting, comparing and classifying the data into themes and patterns (Hsieh & Shannon, 2005). To prepare for the data analysis, the handwritten, populated INRS were transcribed into SPSS and cleaned. The specific process for cleaning, reducing and coding the data was documented in a coding manual and updated throughout the coding process (Miles and Huberman, 1994). For example, the active forms of verbs were used so that “he will cry” was replaced with “cries”. Decisions and rationales for reducing and coding data were documented in the coding manual. In addition, an audit trail documented the process of the study through completion.

The data was reduced and categorized by the PI. In this study, the analysis was non-directed, meaning that the categories for evaluating the frequency counts were not predetermined, but was established during the data analysis (Hsieh & Shannon, 2005). The criteria for analyzing the data were determined as follows. Each parent’s descriptions of their child’s pain responses were read thoroughly. The descriptions were reread and categories of the similar pain descriptions 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
was examined for the emergence of new categories and categories were readjusted and defined. For instance, initially a category of physical activity related to extremities was created. Upon closer examination of the data, I found that the data really described two different responses, muscle tone and physical activity. So this category was divided into categories, “Muscle tone” and “Activity Level”.

Most of the pain cues fit easily into categories. A 3-member panel of experts was consulted to provide judgments on ambiguous pain responses that did not fit easily into one category. The expert panel consisted of nurses with extensive experience assessing post operative pain in nonverbal children with ID. The panel included 3 surgical nurses (2 being bedside nurses on a surgical unit, one being a master’s prepared surgical nurse practitioner). The panel was chosen for their interest in this population and in improving pain assessment in general. The panel’s mean years of nursing experience was 28 years with a range from 26-35 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. The expert panel was given an abstract of the study and a copy of the categorized pain cues. The expert panel was asked to categorize 18 ambiguous pain responses that did not fit easily into one category. An example of an ambiguous pain response is “makes fists”, which without knowing the motive for the behavior could be a social behavior or could be a physiological response of increased muscle tone. In this case, having seen this response when caring for similar children, the expert panel unanimously selected the category, “muscle tone”. The feedback from the panel also resulted in redefining the categories. For example, activity level was clarified to include only activity while awake. So pain
cues that described sleep as a response to pain were included in physiologic and not activity level.

Once the final categories were derived and defined, then frequency counts of the categories were calculated to explore the incidence of these pain responses. Then the relationship between the pain responses (nominal level data) and the following independent variables was examined using chi square statistics. The independent variables collected as nominal data were gender and reason for ID. 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 the chi square analysis. For example, for the chi square analysis, severity of pain was re-grouped from interval (0-10) to categorical data: no pain, mild pain (INRS 1-3), moderate pain (INRS 4-7), and severe pain (INRS 8-10).

The final step in the research study was a comparison of the pain cues identified in the literature and the parent described pain responses collected in this study. The purpose of this analysis was to evaluate the pain assessment tools in the literature to evaluate whether or not these tools were inclusive of pain responses in children with diverse physical and expressive abilities and with a wide range of pain responses,

**Rigor and Validity.**

Lincoln and Guba (1985) translated the attributes of strong quantitative research into concepts more compatible with the philosophical underpinnings of qualitative research. These concepts include credibility (from internal validity), transferability (from external validity), dependability (from reliability) and confirmability (from objectivity).
Below, each of these concepts, credibility, transferability, dependability and confirmability, is discussed in relation to this study.

**Credibility.**

Credibility refers to the truth value of the study. Hsieh & Shannon (2005) emphasized that demonstrating the consistency between interpretation of the data and the actual data, increases credibility. Towards this effort, member checking was used during data collection, for the original research (Lincoln & Guba, 198). In the original research from which data for this study were drawn, parents were given the opportunity to review and edit the populated pain tool before each set of pain assessments completed (1-2 times). This was done in order to confirm the information collected at the initial meeting in the preoperative clinic. In addition, an expert panel, not involved in the original study, validated the analysis of this study by confirming the reduced data.

**Trustworthiness.**

The study of pain responses in this population has been examined primarily through quantitative methods. Looking at the data and results reported in the literature from a qualitative approach will add a different perspective. Results that are complementary, confirm the trustworthiness of the study (Breimayer, Ayres, Knaft, 1993). Also this comparison between the collected parent identified pain responses and pain cues identified in previously published documents allows for a comparison between ‘emic’ and ‘etic’ information. ‘Emic’ refers to a description of responses as described from an observer within a certain culture. This would include the parent identified pain cues. ‘Etic’ refers to a description of a response by an outside observer in this case a team of researchers.
Transferability.

Transferability refers to whether the study results can be applied to different settings or populations. In the original study of the psychometric testing of the INRS, efforts were made to increase the transferability of the study. For example, the parent described pain behaviors were collected in the format of the individualized numeric rating scale. Although the format of the INRS may have limited the space to document the child’s responses, having limited space to document pain responses is more in keeping with clinical practice in a hospital setting, so this may actually increase the chances that the results will be transferable to a hospital setting.

In addition, the size of the sample (n=50) was adequate for psychometric testing and the sample size is large for a qualitative study. Theoretically, a larger sample should contribute fewer sampling errors that occur as a result of random variation.

It could be said that the transferability of the results of this study is limited to this specific population of nonverbal children with ID for this reason. However, if all children, even those with good verbal abilities and mild intellectual disabilities, were included in the sample, the results would be more generalizable to a broad range of children but the results would not be specific enough to apply to nonverbal children with ID. Since the problem of misinterpreting pain cues occurs in nonverbal children with ID and not typically in children with mild ID and good communication skills, efforts were made to focus on the population in need.

Another limitation regarding transferability is related to the study being a secondary data analysis. The inclusion criteria for the original study, children requiring elective surgery with an overnight stay in which the parent is present for part of the
hospital admission, may have limited the sample to include a group of children with more health issues or perhaps with very attentive parents who recognized their child’s pain, pushed for surgical interventions and those parents who have the resources and support to remain at the hospital bedside overnight.

**Dependability.**

Dependability (consistency) was strengthened by a member check in the original research study with parents to verify or make changes to the descriptions of the child’s responses to pain. A coding manual was used as a reference for consistency. This manual documented how words were coded and how the themes evolved. In addition, the use of an expert panel during the analysis also improved the dependability of the study.

Since the author of this study is also the primary author of two previously described articles, written about the INRS, special efforts were made to increase confirmability (neutrality) in this study. Bracketing was used in order to make knowledge or biases overt by writing it down. A diary was used to document assumptions, possible relationships and outcomes throughout the analysis.
Chapter 4: Results

These are the results of a study of parent described pain responses in nonverbal children with ID. This study drew upon parental knowledge of the child through an examination of the words that parents of nonverbal children use to describe their child’s pain cues. The overall purpose of this study was to examine the words that parents of children with ID use to describe their child’s pain responses to identify patterns of pain responses that may be clinically useful during pain assessment. The goal of this study was to better understand the nature of pain expression in this population. The research questions were: What themes or patterns can be identified through an examination of parent reported descriptions of pain cues in nonverbal children with ID? What is the relationship between type of pain responses and the following independent variables: child expressiveness, cause of ID, child gender, child developmental age and severity of pain and child co-morbidities and parent age? In relation to the purpose, goal and questions, the aims were to:

1) Identify common pain responses as described by parental report.

2) Examine the relationship between type of parent described pain responses of children with ID and the following independent variables: child expressiveness, cause of ID, child gender, child developmental age, severity of pain, child co-morbidities, and parent age.

3) Compare the parent identified pain responses to the pain cues described in the literature for this population.

In the context of the purpose, goal and questions, the findings will be discussed in relation to the three aims. First, the demographics of the children and their parents in this study are described, then the findings for each aim.
**Child Demographics**

In the original research from which the data for the secondary analysis study were obtained, a convenience sample of on 50 nonverbal children ages 6-18 years with ID was recruited from the preoperative and the orthopedic clinics. Data was collected between July 2004 and April 2008. The demographics of the sample of children are listed in Table 4. The mean level of functioning was measured by the Vineland Adaptive Behavior Scale in 3 domain scores; Communication (Mean =16.0 months, S.D. 6.5), Daily Living Skills (mean 15.1 months, S.D. 4.9), Socialization (mean 18.7 months, S.D. 8.4). The fourth domain of the VABS, motor skills was not assessed as participants were extremely limited in their independent motor skills. The most common cause of ID reported by the parents included prenatal or perinatal events (n=30) and seizure disorders (n=4). 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%).
Table 4

*Child Demographics (n=50)*

<table>
<thead>
<tr>
<th>Measure</th>
<th>Female</th>
<th>21; 42%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Gender (N; %)</td>
<td>Female</td>
<td>21; 42%</td>
</tr>
<tr>
<td></td>
<td>Male</td>
<td>29; 58%</td>
</tr>
<tr>
<td>Age (Mean; range)</td>
<td></td>
<td>10.4 years; 6-18 years</td>
</tr>
<tr>
<td>Ethnicity (N; %)</td>
<td>Hispanic/Latino</td>
<td>13; 26%</td>
</tr>
<tr>
<td>Race* (N; %)</td>
<td>White</td>
<td>43; 86%</td>
</tr>
<tr>
<td></td>
<td>Black/African American</td>
<td>4; 10%</td>
</tr>
<tr>
<td></td>
<td>Grouped others</td>
<td>2; 4%</td>
</tr>
<tr>
<td>Vineland Adaptive Behavior Scale</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Communication (Mean, S.D.)</td>
<td></td>
<td>16.0 months, S.D. 6.5</td>
</tr>
<tr>
<td>Daily Living Skills (Mean, S.D.)</td>
<td></td>
<td>15.1 months, S.D. 4.9</td>
</tr>
<tr>
<td>Socialization (Mean, S.D.)</td>
<td></td>
<td>18.7 months, S.D. 8.4</td>
</tr>
<tr>
<td>Motor Skills (Mean, S.D.)</td>
<td></td>
<td>Not assessed**</td>
</tr>
</tbody>
</table>

** motor skills were not assessed as participants were extremely limited in their independent motor skills. The additional questions may be burdensome to parents without being of benefit.
Parent Demographics

The parents of the previously described 50 children were interviewed in a semi-structured interview about their child’s pain cues. The parents were mostly female (88%), white (80%) with a mean age of 40 years (range 27-59 years old). 52% of the parents had a high school education and 42% had a college degree (Table 5).

Parents described their child’s pain responses and ranked the responses on a scale of 0-10, 0 being no pain and 10 being worse imaginable pain. In total, 423 pain responses were collected. The figures below detail the total number of pain responses described by the parent(s) (mean 8.4 descriptors) (Figure 1) and the distribution of the pain cues by severity on a scale from 0-10 (Figure 2). Parents reported 53.4% of the pain cues at 3 points along the 0-10 continuum: no pain (n=88, 20.8%); at a pain intensity of 5 (n=57, 13.5%) and a pain intensity of 10 (n=76, 18%) (Figure 2).
Table 5

*Parent Demographics (n=50)*

<table>
<thead>
<tr>
<th>Measure</th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Gender (N; %)</td>
<td>Female</td>
<td>44; 88%</td>
</tr>
<tr>
<td></td>
<td>Male</td>
<td>6; 12%</td>
</tr>
<tr>
<td>Age (mean; range)</td>
<td></td>
<td>40 years; 27-59 years</td>
</tr>
<tr>
<td>Ethnic group</td>
<td>Hispanic/Latino</td>
<td>6; 12%</td>
</tr>
<tr>
<td>Race (N; %)*</td>
<td>White</td>
<td>40; 80%</td>
</tr>
<tr>
<td></td>
<td>Black/African American</td>
<td>4; 8%</td>
</tr>
<tr>
<td></td>
<td>Other</td>
<td>2; 4%</td>
</tr>
<tr>
<td>Primary Language</td>
<td>English</td>
<td>42; 84%</td>
</tr>
<tr>
<td></td>
<td>Spanish</td>
<td>4; 8%</td>
</tr>
<tr>
<td></td>
<td>Other</td>
<td>2; 2.4%</td>
</tr>
<tr>
<td>Highest Education</td>
<td>Less than high school</td>
<td>2; 4%</td>
</tr>
<tr>
<td></td>
<td>High School Graduate</td>
<td>26; 52%</td>
</tr>
<tr>
<td></td>
<td>College Graduate</td>
<td>15; 30%</td>
</tr>
<tr>
<td></td>
<td>Graduate education</td>
<td>6; 12%</td>
</tr>
</tbody>
</table>
Figure 1

Number of Parent Described Pain Cues per Patient

Number of Indicators used by parents to describe child’s pain response
Figure 2

*Distribution of Parent Described Pain Cues by Severity (on a scale from 0-10)*
Results Aim #1: Categorized Parent Described Pain Responses

The 423 parental described pain responses were transcribed into SPSS, cleaned and reviewed. Of the 423 parent described pain cues, 88 described when the child had no pain or was in their usual state. Although these baseline behaviors are important to have when assessing patients for pain, they were not included in the analysis of pain cues since these cues do not describe pain cues. However, examination of these ‘no pain cues’ (n=88) such as “not vocalizing”, “active, moves a lot”, “seeks physical closeness”, “moans” or “sleeps” found that 25% (n=22) of the no pain cues were descriptors of pain in other children.

The criteria for analyzing the remaining 335 pain cues were determined during the data analysis. Most of the pain cues fit easily into 7 distinct categories of pain responses: 1) vocalizations including crying; 2) social behaviors; 3) facial expression; 4) physiologic measures; 5) muscle tone; 6) activity level and; 7) self-injurious behaviors. An expert panel reviewed the categorized pain cues and helped to categorize 18 ambiguous pain responses that did not fit easily into one category. An example of an ambiguous pain response is “makes fists”, which could be a social behavior or could be a physiological response of increased muscle tone. In this case, having seen this response when caring for similar children, the expert panel unanimously selected the category, “muscle tone”.

The greatest percentage of pain cues were clustered within the categories of vocalizations including crying (39.4%), followed by facial expression (21.8%) and social behaviors (16%). Vocalizations included a variety of vocal responses including yelling, screaming and crying. It also included not just vocal responses but a change in
vocalizations. For example, some children were quite vocal at baseline and parents described a change in vocalizations from vocal to quiet as a pain response.

Social behaviors included a range of behaviors from reaching out for comfort to withdrawing from social contact. Examples include “withdraws from other” and “tries to hide or bury self”.

Facial expression included changes that manifested within the face including changes in the eyes, mouth or general facial expressions. Examples of facial pain responses include “eyes look scared” and “eyes look glassy”.

Physiologic changes included pain responses such as changes in heart rate, blood pressure, changes in breathing pattern and changes in skin color, “looking sick”. Eating and sleeping was initially coded as a separate category but was eventually combined with physiologic after discussion with members of expert panel.

Muscle tone also included a wide range of behaviors from floppy to tight muscle tone. Examples include “arms floppy” and “arms tighten up”.

Activity level, for the purposes of this study, only included pain responses while the patient was awake. The feedback from the panel helped to clarify this category. Pain cues that described changes in sleep patterns were categorized as physiologic and not activity level. Within the category of activity level, pain responses ranged from increased activity to little activity.

Within this sample of 50 children, only 8 (16%) had self-injurious behaviors in response to pain. Self-injurious behaviors included pain responses such as “pulls on eyelids”, “bites hands”, “bites lips” or “hits head”.
Within four of the categories, vocalization, social behaviors, muscle tone and activity level, the pain cues had a wide range of responses, and even included disparate responses to pain. For example, as noted within the category of vocalization/crying, some parents described increased vocalizations and some described decreased vocalizations from the child’s usual baseline. With the category of social cues, some children responded to pain by withdrawing from human contact and others reached out for human comfort. Similarly within the muscle tone category, some parents described increased muscle tone and others described decreased muscle tone. Within the activity level category, some children exhibited increased activity when in pain while others exhibited decreased activity level when in pain. Table 6 outlines the results of the summative qualitative content analysis including categories and examples.
Table 6

*Categorized Pain Responses: Frequencies and Examples*

<table>
<thead>
<tr>
<th>Categories</th>
<th>n</th>
<th>%</th>
<th>Examples</th>
</tr>
</thead>
<tbody>
<tr>
<td>Vocalizations including Crying</td>
<td>132</td>
<td>39.4%</td>
<td>Vocalizing stops</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Snorting noises</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Whimpering</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Occasional growl</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Crying</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Screams</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Howling</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Yelling angry sounds</td>
</tr>
<tr>
<td>Social Behaviors</td>
<td>73</td>
<td>21.8%</td>
<td>Tries to hide or bury self</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Fitful with strangers</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Withdraws with arms over head</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Holds people hands</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Seeks physical closeness</td>
</tr>
<tr>
<td>Facial Expression</td>
<td>53</td>
<td>16.0%</td>
<td>Wincs</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Looks concerned</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Eyes look scared</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Eyes squinting</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Eyes look glassy</td>
</tr>
</tbody>
</table>
Table 6

*Categorized Pain Responses: Frequencies and Examples (Continued)*

<table>
<thead>
<tr>
<th>Categories</th>
<th>n</th>
<th>%</th>
<th>Examples</th>
</tr>
</thead>
<tbody>
<tr>
<td>Physiologic</td>
<td>24</td>
<td>7.2%</td>
<td>Heavy breathing</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Seizes with extreme pain</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Respiratory rate high</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Sweats</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Sleeps</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Red in the face</td>
</tr>
<tr>
<td>Muscle Tone</td>
<td>23</td>
<td>6.9%</td>
<td>Arms floppy</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Extends arms, tense arms</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Makes fists</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Grinds teeth</td>
</tr>
<tr>
<td>Activity Level</td>
<td>20</td>
<td>6.0%</td>
<td>Starts to look very tired</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>More physically active</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Kicks</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Thrashes around bed</td>
</tr>
<tr>
<td>Self -injurious</td>
<td>9</td>
<td>2.7%</td>
<td>Bites on hand</td>
</tr>
<tr>
<td>Behaviors</td>
<td></td>
<td></td>
<td>Pulls on eyelids</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Self abusive in different ways</td>
</tr>
<tr>
<td>Total</td>
<td>335</td>
<td>100%</td>
<td></td>
</tr>
</tbody>
</table>
Results Aim #2: Relationship between Pain Response and other Variables

Once the pain cues were categorized, the relationships between parent described pain responses (categories listed in Table 6) and certain independent variables (listed in Table 7) were explored using chi square statistics. The relationships between the children’s characteristics and categories of pain responses were examined to determine if these independent variables were related to the child’s pain responses. The relationships between the parent characteristics and categories of pain responses were explored to determine if these variables were related to the way parent’s described their child’s pain. Variables that were collected as interval variables were regrouped into meaningful grouping as nominal variables for the chi square analysis. For example, as noted, severity of pain was collected as interval data from 0, meaning no pain, through 10 meaning worst possible pain. For the chi square analysis, severity of pain was regrouped as no pain (0), mild pain (1-3), moderate pain (4-6), severe pain (7-10).

Severity of pain was significantly related to the categories of pain responses, \[ \chi^2_{21, n=423} = 451.13, \ p < .001 \]. Parents reported changes in the child’s facial expression more often when describing mild to moderate pain responses. Parents reported changes in vocalizations more often when severity of pain intensity increased. Vocalizations increased incrementally as pain severity increased (Figure 3). Physiologic responses to pain were described more often during moderate to severe pain. Surprisingly, parents described fewer changes in facial expression as pain severity increased. Self-injurious behavior and social behavior did not increase as pain severity increased.
Cause of ID was collected by reviewing the medical record and if needed, clarifying this information with the parent. Cause of ID was significantly related to the pain categories \[X^2 (14, n=423) =38.79, p=<.000\] with more changes in vocalizations and more self injurious behaviors being reported in children with seizure disorders.

The relationship between gender of the child and categories of pain response was significant \[X^2 (7, n=423) =18.62, p=<.01\]. Parent’s reported more physiologic and social cues in daughters and more changes in vocalization and fewer physiologic and social changes in sons than statistically expected in the chi square analysis. The number of the child’s co-morbidities, parent gender, parent age, 3 domains of the VABS: child communication ability, daily living skills and social skills and child developmental age (total score on the VABS) were not significant in this analysis (Table 7).
Figure 3

Categories of Pain Response by Severity

Frequency

Categories of Pain

Pain Severity
Table 7

*Chi Square Analysis: Pain Response and Independent Variables (n=423)*

<table>
<thead>
<tr>
<th>Independent Variables</th>
<th>df</th>
<th>$X^2$</th>
<th>P value</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Child Characteristics</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<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 gender</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>
</tr>
<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>
</tr>
<tr>
<td><strong>Parent Characteristics</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Parent gender</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>
</tr>
</tbody>
</table>

*df* degrees of freedom

*$X^2$ chi square statistics

*ID* intellectual disability

* significance
Results Aim #3: Comparison of Pain Responses to Published Pain Assessment Tools

In order to examine the comprehensiveness of the pain assessment tools for this population, each of the 335 parent described pain responses was categorized according to the items described in pain assessment tools. The pain assessment tools included in this analysis were: r-FLACC, NCCPC, PICIC, PPP and UWCH. The INRS was not evaluated in this analysis because items used in this study were obtained from this tool. Thus all items would be included. Also, the flexible nature of the INRS allows for child specific pain indicators.

Each of the tools had outliers of the 335 pain responses that did not fit into the pain tools. Of the 5 tools examined, the NCCPC was the most comprehensive. Only 2.8% (n=12) of the 335 pain responses were not included within the tool’s items. Both the UWCH and the PPP 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 8). For each of the 5 tools, crying (though defined differently for each tool) was the highest frequency category.

In summary, the results detail the wide range of pain responses in this population within 7 common categories. The results describe the relationships between the 7 categories of pain responses and 3 independent variables: gender, pain severity and cause of ID. For the wide range of pain responses in this population of children, the INRS and the NCCPC are the most comprehensive pain assessment tools when compared with the parent described pain responses of this study.
Table 8

*Parent Described Pain Responses and Items of Pain Assessment Tools*

<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”</td>
<td>“Crying”</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Increased HR</td>
<td>0.5% (n=2)</td>
<td>27.9% (n=118)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Sweats</td>
<td>(Includes all vocalizations-can individualize)</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Increased BP</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Increased temperature</td>
<td></td>
<td></td>
</tr>
<tr>
<td>NCCPC</td>
<td>30</td>
<td>2.8% (n=12)</td>
<td>Heart rate high</td>
<td>“Shivering”</td>
<td>“Cry”</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Increased BP</td>
<td>0% (n=0)</td>
<td>15.8% (n=67)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Increased temperature</td>
<td>(Separate category for “special sound or vocalization”)</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Vocalization stops</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Stops crying</td>
<td></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”</td>
<td>“Crying”</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Vocalizations stop</td>
<td>moves away</td>
<td>17%</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Clapping hands</td>
<td>when (n=72)</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Laughing silly</td>
<td>touched”</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Stops crying</td>
<td>4.7% (n=20)</td>
<td></td>
</tr>
</tbody>
</table>
Table 8

*Parent Described Pain Responses and Items of Pain Assessment Tools (Continued)*

<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>PPP</td>
<td>20</td>
<td>31.9% (n=135)</td>
<td>Sweats</td>
<td>“Flexed”</td>
<td>“Crying,”</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Heavy breathing</td>
<td>inward or</td>
<td>moaned,</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Seeks comfort by</td>
<td>drew legs</td>
<td>“groaned”</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>physical closeness</td>
<td>upwards</td>
<td>26.7% (n=113)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Increased HR</td>
<td>towards</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Vocalizations stop</td>
<td>chest”</td>
<td>0.2% (n=1)</td>
</tr>
<tr>
<td>UWCH</td>
<td>5</td>
<td>31.9% (n=135)</td>
<td>Arms floppy</td>
<td>“Sleep”</td>
<td>“Vocal/Cry”</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Stops eating</td>
<td>0.5% (n=2)</td>
<td>23.2% (n=98)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Increased seizures</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Respiratory rate high</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Vocalizations stop</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
Chapter 5: Discussion

This is a discussion of the results of a study of parent described pain responses in nonverbal children with ID. The study drew upon parental knowledge of the child through an examination of the words that parents of nonverbal children use to describe their child’s pain cues. The overall purpose of this study was to examine the words that parents of children with ID use to describe their child’s pain responses to identify patterns of pain responses that may be clinically useful during pain assessment. The goal of this study was to better understand the nature of pain responses in this population. The research questions were: What themes or patterns can be identified through an examination of parent reported descriptions of pain cues in nonverbal children with ID? What is the relationship between type of pain responses and the following independent variables: child expressiveness, cause of ID, child gender, child developmental age and severity of pain and child co-morbidities and parent age? In relation to the purpose, goal and question, the aims were to:

1. Identify common pain responses as described by parental report.
2. Examine the relationship between type of parent described pain responses of children with ID and the following independent variables: child expressiveness, cause of ID, child gender, child developmental age, severity of pain, child co-morbidities, and parent age.
3. Compare these parent identified pain responses to the pain cues described in the literature for this population.

Accurate pain assessment of nonverbal children with ID is crucial because these children are at risk for having pain and for having pain that is misunderstood. Despite
several published standardized pain assessment tools, at this time, none of the standardized pain assessment tools meets the requirements of a well established pain assessment tool as defined by Cohen, Greca, Blount, Kazak, Holmbeck, Lemanek, 2008. All of these tools require further testing. In addition to further testing, a better understanding of the nature of pain responses in this population is needed in order to judge the clinical utility of pain assessment tools.

This chapter is structured as follows. First, the results of the 3 aims are discussed. This is followed by study limitations, implications for practice, research, education and policy.

**Discussion Aim #1: Categorized Parent Described Pain Responses**

The first aim was to identify common pain responses as reported by parents. All 50 parents in the study were able to complete the task of describing their child’s pain cues (when in pain and when not). In addition, all parents were able to rank order these descriptors and link them to the numeric rating scale for pain.

The task of rank ordering the child’s pain behaviors adds to the literature by documenting that parents are able to articulate their child’s pain behaviors and are able classify differences as exhibited during mild, moderate or severe pain. Only one parent of an autistic boy had more difficulty articulating her son’s pain behaviors and had fewer pain descriptors. This parent reported that her son did not always respond consistently to similar painful stimuli such as receiving immunizations at the pediatrician’s office. These findings support the common clinical practice of relying on parent’s assessments of pain (Herr et al, 2006) especially without a well established pain assessment tool for children with intellectual ability (Cohen et al, 2008).
The results of the summative content analysis found 7 distinct categories of pain behaviors: 1) vocalizations including crying; 2) social behaviors; 3) facial expression; 4) physiologic measures; 5) muscle tone; 6) activity level and; 7) self-injurious behaviors. These results support statements in the literature that pain responses in this population are ‘individual’ or ‘idiosyncratic’ (McGrath et al, 1998; Terstegen et al, 2003; Carter et al, 2002; Fanurik et al, 1999). These results are similar to the 5 categories of the Non-Communicating Children’s Pain Checklist (NCCPC): 1) Vocal; 2) Social; 3) Facial; 4) Activity; 5) Physiologic (McGrath, 1998); the 6 categories described in Terstegen (2003): (1) facial expression; (2) motor behavior; (3) social behavior/mood; (4) attitude toward sore body part; (5) vocalization; (6) physiological signs; and the 5 categories of the FLACC: 1) Face; 2) Legs; 3) Activity; 4) Cry and; 5) Consolability (Merkel, 1997).

Interestingly, as noted in the findings, parents reported the majority (53.4%) of their child’s pain cues at 3 points along the 0-10 continuum of the INRS: no pain (n=88, 20.8%); at a pain intensity of 5 (n=57, 13.5%) and a pain intensity of 10 (n=76, 18%). These results warrant further investigation as to whether the INRS could be simplified from 11 points to document to perhaps 4 (no pain, mild pain, moderate pain, severe pain).

Most of the parent described pain cues in this sample, fell into 3 categories: vocalization including crying (39.4%), social behaviors (21.8%), and facial expression (16%). This supports previous evidence that changes in facial expression are an important part of pain assessment in this population (Stallard et al, 2002; Breau et al, 2002; Voepel-Lewis et al, 2002; Carter et al, 2002). However in this study, vocalizations including crying (39.4% of pain cues reported in this study) and social behaviors (21.8% of pain
cues reported in this study) were more frequently reported by parents than facial expression (16% of pain cues reported in this study).

Despite the documented common categories of pain responses, nonverbal children with ID express pain within a range of observable cues. For example, as noted in the findings, some children express pain through increasing vocalizations, while others express pain through a change in vocalization or even when vocalization stops. Some children express pain by withdrawing from social contact, while others seek physical closeness. Some children respond to pain with decreased muscle tone and become “floppy”, while others have increased muscle tone and become spastic. Some children have increased activity level with pain, while others become very still and do not move.

Divergent pain expressions in this population have important clinical implications. Creating a pain assessment tool for clinical use that captures these extremes in pain response is challenging. The NCCPC includes the extremes in pain response as separate items on the pain assessment tool so that each extreme is included. Including each of the extremes of pain such as “body/limbs floppy” and “spastic tense rigid”; “less interaction, withdrawn”; “not moving, less active, quiet” and “jumping around agitated, fidgety” improves the comprehensiveness of the NCCPC. However, the problem with including extremes of pain response as separate items in a pain assessment tool is that if a child scores high in one item then the child would score low in the opposite extreme. 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.

Both the INRS and the r-FLACC allows for extremes in pain response by individualizing the pain assessment tool to the child’s past responses to pain. However,
the 5 item structure of the r-FLACC, limits its comprehensiveness to only the 5
categories of pain response (Face, Legs, Activity, Cry and Consolability), while the
structure of the INRS is more accommodating to a variety of pain responses including
behavioral as well as physiologic responses to pain.

Discussion Aim #2: Relationship between Pain Response and other Variables

Differentiating Pain Severity

It is a common assumption that a child will exhibit a greater number of pain cues
and a greater expression of certain pain cues as pain intensity increases. The current pain
tools published in the literature, with the exception of the Individualized Numeric Rating
Scale (INRS), are based on this assumption. For example, within the Face Category of
the FLACC, the descriptions depict increasing facial expression as pain intensity
increases. For example, “No particular expression or smile” describes the lowest score in
the Face Category. This is followed by “Occasional grimace/frown, withdrawn or
disinterested” then by “Frequent/constant quivering chin, clenched jaw” (Merkel,
Voepel-Lewis, Shayevitz, Malviya, 1997). The other categories within the FLACC
follow this assumption in a similar manner. The results of this study contradict this
assumption.

In this study, there was a significant relationship between categories of pain and
pain severity using a chi square analysis. As pain intensity increased, the type of pain
response changed. For example, a child may express mild pain by grimacing but a change
in muscle tone may indicate severe pain. Only the category “changes in vocalization
including crying” follow the common belief and increase in number as the severity of
pain increases. In contrast, the following pain cues: physiologic cues, changes in muscle
tone, and changes in social behaviors, peaked at moderate pain. Interestingly, the parents in this study reported more changes in facial expression with mild and moderate pain than with severe pain.

There are several possible explanations for parents reporting less facial expression during severe pain. It may be that children exhibit fewer facial cues when in severe pain. Another explanation is that during severe pain, parents may be more attentive to apparent changes such as vocalizations or muscle tone. A third explanation is that the child’s escalating vocalizations, being so easy to recognize, obscures the parent’s recall of facial changes during severe pain.

Three categories (physiologic cues, changes in muscle tone, and changes in social behaviors) were found to increase from mild to moderate pain and to peak at moderate pain. An explanation for this finding may be that this population of children has physical limitations that make it more difficult for them to express severe pain. In this sample, self-injurious behaviors remained steady regardless of pain intensity. However, the sample had too few patients who exhibited self injurious behaviors to presume any pattern. Regardless of the reasons, these results give preliminary evidence that pain intensity in this population is not necessarily best expressed by the total number of pain cues. These results have implications for pain assessment tool development for this population. An individualized approach based on knowledge of the child’s pain responses may be the most accurate pain assessment for this population.

**Cause of Intellectual Disability**

As previously mentioned, nonverbal children with ID are a diverse group of children with different physical and expressive abilities. Intellectual disability occurs as a
result of many different physical conditions, such as prenatal events, post natal infection, chromosomal abnormalities or even accidents such as near drowning or head injuries. The sample in this study was categorized into only 3 distinct groups with different causes of ID: prenatal insult, seizure disorder and a category of children with “other” causes of ID including near drowning and chromosomal disabilities. The causes of ID in this “other” group occurred too infrequently to characterize as a single group.

Chi square statistics were used to measure relationships between types of pain responses and some selected child characteristics as independent variables. There was a significant relationship between pain response and cause of ID (p<.001). Children whose cause of ID resulted from a prenatal insult expressed pain using more facial expressions and fewer social and vocal cues than expected in the analysis. Children whose cause of ID resulted from a seizure disorder used more physiologic and social cues and fewer facial cues than expected. Since the cause of ID directly affects the child’s physical abilities and in some situations the source of pain, it follows logically that children with similar causes of ID would respond similarly to pain. Further study with a larger sample size and more varied causes of ID is needed to determine if the cause of ID could help to predict pain responses in other children.

**Role of Sex and Gender**

Over the last 20 years, there has been a growing body of knowledge about the role of sex, gender and pain (Hurley & Adams, 2008). According to the Institute of Medicine, Exploring the Biological Contributions to Human Health: Does sex matter?, “sex” is the classification of living things, generally as male or female according to their reproductive organs and function assigned by chromosomal complement”. Gender is defined as “a
person’s self representation as male or female or how that person is responded to by social institution on the basis of the individual’s gender presentation” (Wizeman & Pardue, 2001). Within this same publication, three reoccurring themes were outlined from research: 1) being male or female is an important basic human variable; 2) the study of sex differences is evolving into a mature science and; 3) barriers to the advancement of knowledge about sex differences exist and must be eliminated (Wizeman & Pardue, 2001).

In this study, there was a significant relationship between the 7 categories of pain responses and sex or gender of the child \[X^2 (7, n=423) =18.62, p=.01\]. Parent’s reported more physiologic and social cues in daughters and more changes in vocalization and fewer physiologic and social changes in sons than statistically expected in the chi square analysis.

There are several possibilities for these differences between males and females. It may be explained by sex differences. Differences in pain thresholds in male and female rodents have been documented, with female rodents having a lower pain threshold than males (Sternberg, Smith, Scorr 2004; Chesler, Wilson, Larivere, Rodriguez, 2002). If this evidence is applicable to humans and to children with ID, one might predict that males, having a higher pain threshold than females, would have more physiologic cues of pain. So males would endure a greater pain stimulus before expressing pain than females. A greater pain stimulus would likely cause physiologic changes. However, the results of this study found that parents reported fewer physiologic cues in their sons as compared to daughters.
The differences in males and females may be explained by gender differences. A child may learn to express pain differently based on parental acceptance and response to certain gender specific pain expressions. 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 may perceive their daughters pain responses differently than their son’s pain responses. It is possible that fathers may perceive their daughters and son’s pain responses differently than mothers. The chi square analysis did not find any relationship between parent gender and categories of pain responses. However, the number of fathers in the study was not adequate to compare mothers’ and fathers’ perceptions of pain responses.

Continued research is needed to further understand the impact of sex, gender and pain responses in this population. However, the literature suggests that the differences between pain expression in males and females are likely explained by gender (of the parent or child) rather than sex differences. Exploration into the impact of social conditioning, social and cultural influences on pain responses is needed especially since this population is greatly influenced by their caregivers.

In addition to parent gender, the relationships between pain responses and the independent variables of child co-morbidities, child developmental age, child communication ability and parent age, were not significant. The lack of a significant relationship between categories of pain and 3 of these variables (child co-morbidities, child development age, and child communication ability) may be explained by the sample. The inclusion and exclusion criteria only permitted nonverbal children with ID who required full physical support of caregivers into the study. For this reason, the
demographics of the sample had a narrow range within these variables. For example, children in this sample had a narrow range of co-morbidities, developmental ages, and child communication abilities.

**Discussion Aim #3: Comparison of Responses to Published Pain Assessment Tools**

Another challenge for pain assessment tools is the balance being comprehensive and concise. The comprehensiveness of a pain assessment tool contributes to the accuracy of the tool. Being concise contributes to the ease of use. The NCCPC-PV was the most comprehensive of the standardized tools. Although other tools were more concise than the NCCPC-PV they did not account for at best 17% (r-FLACC), at worst 32% (PPP and UWCH) of the parent described pain cues when comparing the items on the pain assessment tools with the parent described pain cues.

Another challenge of pain assessment in this population is that some of the parent described cues for ‘no pain’ are cues of pain in other children. In this study 25% (n=22) of the no pain cues (n=88), such as “not vocalizing” or “active, moves a lot”, “seeks physical closeness”, “moans” or “sleeps”, were descriptors of pain in other children. Both findings (1) the individual responses to pain within general categories of pain responses and; 2) that ‘no pain’ descriptors are cues of pain in other children) support the use of knowledge of the individual child to aid in pain assessment in this population. Two pain assessment tools, the INRS and the PPP allow for documentation of baseline behaviors for comparison during pain assessment.

The results of the summative, qualitative content analysis found that physiologic cues of pain comprised only 7.2% of the pain cues described by parents. It may be that the numbers of physiologic cues of pain are low because parents do not recognize these
cues. Parents may not have easy access to physiologic indicators of pain such as heart rate, respiratory rate, blood pressure, oxygen saturation. Or perhaps the low percentage of physiologic pain cues reported by parents is because this population of children often has pain daily and the body has accommodated to this pain, therefore physiologic changes are subtle or not present. Another explanation is that parents understand that vital sign changes do not often differentiate between pain and distress as noted by other researchers on pain (McCaffery & Pasero, 1999). Physiologic cues of pain require further investigation to determine the importance of these cues in pain assessment in this population.

**Limitations**

There were several methodological limitations noted in this study. These limitations are: 1) the sole use of parent knowledge of the child for pain cues; 2) related to sampling of parents and children and; 2) related to the study being a secondary analysis.

**Parent Knowledge of Child Pain Cues**

The sole use of parent described pain responses in children with ID without clinician input is a limitation of the study. Including clinicians observations of pain response would strengthen the study. However, the decision to use parent described pain cues for this study is supported by the literature that parents of children with ID know their child’s pain cues (Solodiuk et al, 2010; Voepel-Lewis et al, 2005; Terstegen, et al, 2003; Hunt, et al, 2003; Solodiuk & Curley, 2003; Carter et al, 2002; Fanurik et al, 1999; McGrath et al, 1998) and years of clinical observations.
**Sampling Limitations.**

This sample was a convenience sample collected over a 4 year period (2004-2008). During this time, articles about pain assessment in children with intellectual disabilities were being published. Because of this, the later data collection may have been influenced by the articles.

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. There was no statistical difference in the demographics of the children and their parents who completed the study as compared to those who were enrolled but did not complete the study. The most common reason for not completing the primary study after enrollment was that the children remained intubated and sedated after surgery.

**Limitations of Secondary Analyses.**

As with all secondary analyses, the aims and research questions were limited by the data collected. For example, within several of these categories, more information from the parents could have been elicited to help clinicians to more readily recognize the parent described pain expressions. For example, within the Social Behavior Category, specifics about how the child “seeks physical closeness” would be important to elicit. Examples of parent described pain expressions needing more specifics within the Facial Expression Category are: “looks concerned” and “looks scared”. In this case, sufficient data was collected to examine the 3 aims of the study, but other research questions had to be deferred to future research.
An important strength of this analysis is that the PI for both the initial study and the secondary analysis were the same person, so the purpose and aims were consistent. The data collection and analysis was thorough and complete.

**Implications for Research, Practice and Education**

There are several implications for future directions. These future directions are related to research, practice and education.

**Research.**

At this time, none of the pain assessment tools for nonverbal children with intellectual disabilities currently meet the standard of a well-established pain assessment tool (Cohen et al, 2008). More research on each of the previously described pain assessment tools for this population is needed. In addition to the recommendations for future research outlined in the Review of the Literature, the results of this study stimulated several questions for further study.

Further research is needed to evaluate the practical aspects of how individual differences in pain responses can be expressed easily and concisely for clinical use. A thorough evaluation of the clinical utility of different pain assessment tools for this population would include: ease of use and burden for clinician and parents and techniques to record a child’s individual pain expressions in the electronic medical record.

Understanding the relationship between the expression of pain and the cause of ID (for example, chromosomal abnormalities or prenatal events such as infections or maternal alcoholism or prematurity) is an important area for further investigation. This
understanding would help clinicians to better recognize and interpret pain responses in order to determine the best treatment for the pain.

Further investigation is needed to evaluate whether pain assessment tools tested in acute pain are accurate to assess chronic pain intensity. This is important because this population of children is at risk for having chronic pain from medical conditions as well as acute pain from procedures, surgeries and routine care.

Lastly, investigation into the relationship between sex, gender and pain responses in nonverbal children with ID is important to understand in order to enhance understanding and to avoid misinterpretation of pain expressions. Comparing the pain cues of fathers and mothers of nonverbal children with ID would enhance understanding in this area.

Practice.

While awaiting further research, several findings can be applied to clinical practice. As previously mentioned, not all of the pain assessment tools described in the literature are able to capture the range of pain cues and differences in baseline behaviors as described by parents of nonverbal children with ID. Therefore, evaluate pain assessment tools carefully based on the population served, the psychometric properties of the tool, the comprehensiveness of the tool, and clinical utility. For comprehensiveness, it is advisable to choose a tool that assesses at least 75% of the pain expressions of the population being served.

As compared to the other tools for this population, the INRS has several distinct advantages. The INRS encourages documentation of baseline or no pain behavior; allows the people who know the child to individualize the pain assessment based on the child’s
past responses to pain without the structural limitations of standardized pain assessment tools and allows for weighting of certain behaviors that may differentiate severity of pain.

The FLACC or r-FLACC, being familiar to clinicians, may be judged to have better clinical utility. However, this tool has several limitations for pain assessment for this population. Though, the individualization of the r-FLACC allows the tool to have greater comprehensiveness, the 5-item structure of the tool limits the weight of any of the individualized items to 20%. Also, the r-FLACC does not allow for individualization of items that do not fit into the 5 item structure such as physiologic items, some of the muscle tone responses and self injurious behaviors.

Of the standardized pain assessment tools for this population (not including the INRS), the results of this study found the NCCPC-PV to be the most comprehensive. For this reason, this author recommends the use of the NCCPC-PV when caring for a nonverbal patient with ID in which the parents are unable to be objective about the child’s pain cues, as is the case with the rare but serious clinical issues of Munchausen by Proxy. However, further study on the NCCPC-PV is needed especially to determine how to best assess items that describe extremes or opposites of pain responses.

Considering these strengths and weaknesses and while awaiting further study, this author recommends the use of the INRS using the pain categories and examples listed in Table 6 to help parents populate the INRS. Although some children may have pain cues not included in the table, the list may help parents recall and articulate their child’s pain cues. The exception to this is in cases when the parents are unable to be objective as mentioned above.
Education.

Despite the lack of a well established pain assessment tool, there are several important findings for clinical practice and the education of clinicians and parents of children with intellectual disabilities. First, when self report measures of pain are not appropriate, as is the case with nonverbal children with ID, it should be emphasized to nurses and nursing students to use the parents’ knowledge of the child to assist with pain assessment. Second, a pain cue in one child may be an indicator of no pain for another child. For this reason, it is preferable for pain assessment tools for this population to include baseline behaviors described by someone who knows the child well. Third, 3 categories of pain responses are especially important to focus on because they comprise the majority of pain responses in this population. The categories are: 1) vocalizations including crying; 2) social behaviors and; 3) facial expressions.

Conclusion

Until recently, little research has been published on understanding pain expression in nonverbal children with intellectual disability. This study expands on current knowledge in several ways. First, these results are congruent with Kathryn Barnard’s Child Health Assessment Model (Barnard, 1976) and the published evidence that parents of nonverbal children with intellectual disabilities can articulate their child’s pain responses. This study contrasts with the trend to use standardized pain assessment tools rather than information generated from parent knowledge of the child.

Second, within the general categories of vocalization, social behaviors, muscle tone and activity level, parents describe a range of pain cues, some of which include extremes or opposite pain responses. Third, it corroborates the evidence that changes in
vocalizations and facial expression are the most commonly identified pain cues in this population. Fourth, the results of this study provide preliminary evidence of the relationship between type of pain responses and severity of pain, cause of ID, and child gender.
References


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