PAIN EXPRESSION IN CHILDREN WITH AUTISM SPECTRUM DISORDER (ASD): A FOUNDATION FOR INSTRUMENT DEVELOPMENT

By

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To my parents, for their constant love and encouragement, and for instilling in me the importance of higher education; To my sister, who has the most generous heart, and teaches me how to live and love life; To my brother, whose own academic excellence challenges me daily, and inspired me to persevere these past years; And to my husband, for his unconditional love, and for steadfastly pointing me towards Christ when I need strength
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Little is known about how children with autism spectrum disorder (ASD) experience and express pain. Communication impairments frequently render children with ASD unable to reliably self-report pain, social impairments limit their understanding of interpersonal interaction (and possibly alter the social context surrounding the pain experience), and perseverative and/or repetitive displays of behavior may influence the pain indicators traditionally associated with children who are physically hurting. Additionally, reports of children with autism having a “high tolerance for pain” create confusion and generate misunderstandings regarding whether they are capable of feeling pain.

The overall purpose of this research was to explore how children with ASD express pain, and determine whether it is feasible to develop a pain assessment tool specifically for this population. In order to approach this global research question, a series of two studies were conducted to 1) explore caregiver beliefs about the pain experience in their children with ASD, 2) identify a number of potential pain indicators in this population, and 3) determine which pain indicators are believed by experts to best indicate pain in children with ASD.
Findings revealed that pain is a serious concern among caregivers. Parents struggle to determine when their children are in pain. Study 1 results indicated that children with ASD are capable of experiencing and expressing pain, however their pain expression is unique. Study 2 validated this uniqueness by identifying pain indicators specific to children with ASD, and by demonstrating that indicators relevant to other, similar populations of children are not considered to be relevant in this population. This research supports the initial belief that a pain assessment tool specifically for use in ASD is warranted. Future research will build upon these studies to continue development of a valid and reliable instrument to assess pain in individuals with ASD. Accurate pain assessment is necessary in order to ensure that pain in this population does not go unrecognized, and thus untreated.
In 1989, McCaffery and Beebe established what is now one of the most widely recognized definitions of pain. Their definition emphasizes the subjective nature of the pain experience, and proposes that pain is “whatever the patient says it is and occurs whenever the patient says it does” (p. 7). Given this well established definition, coupled with other nationally acknowledged definitions, the accepted “gold standard” for pain assessment and measurement is self-report. This measurement captures the subjective aspects of the pain experience, and supports the prevailing belief that pain is a unique and individualized process, heavily influenced by the perceptions of those experiencing it. This definition however, inherently implies that the person experiencing pain can verbally communicate his or her subjective experience. The International Association for the Study of Pain (IASP) defines pain as “as unpleasant sensory and emotional experience associated with actual or potential tissue damage, or described in terms of such damage” (IASP, 2007, ¶ 17). This definition also emphasizes the subjective nature of pain, and supports the use of self-report, which solicits how an individual would describe his or her pain to others (Merskey, 1991).

In 1996, Anand and Craig critiqued the most commonly cited pain definitions and stated that “pain assessment must be designed to conform to the communication capabilities of the suffering person” (p. 3). They emphasized the responsibility of scientists and clinicians to adapt a working definition of pain that would include certain vulnerable populations. Specifically, they referred to a) individuals unable to communicate verbally (i.e., infants, or perhaps children and adults with communication impairment), and b) individuals whose verbal self-report might be unreliable (i.e., younger children, and persons with cognitive impairment or dementia). Effective pain assessment and measurement in a variety of populations requires that one
understand how to identify and interpret specific nonverbal indicators of pain, especially when verbal pain expression is confounded by an underlying condition (Oberlander & Craig, 2002).

This research focused on one particular vulnerable population—one that poses an extremely unique challenge to pain assessment—children diagnosed with an autism spectrum disorder (ASD). Communication impairments frequently render children with ASD unable to reliably self-report pain, social impairments limit their understanding of interpersonal interaction (and possibly alter the social context surrounding the pain experience), and unusual (perseverative and/or repetitive) displays of behavior may influence the pain indicators traditionally associated with children who are physically hurting. Additionally, reports of children with autism having a “high tolerance for pain” create confusion and generate misunderstandings regarding whether they are capable of feeling pain. If individuals, specifically healthcare providers, assume that children with ASD are not capable of feeling pain as a result of their underlying disorder, these children are at tremendous risk for having their pain go unrecognized and untreated. At present, it is unclear how children on the autism spectrum cognitively perceive, apply meaning to, and commonly express their physical discomfort.

**Background and Significance of the Problem**

Prior to 1989, the pain experience in children was severely underestimated, if even acknowledged at all. The prevailing belief was that “young children did not feel pain in the same way as adults” (Finley & McGrath, 1998, p. 1). It was also believed that if they do feel pain, their inability to remember the painful event negates the need for astute pain management. The seminal work of Anand (1989) not only established that children, infants, and neonates have fully functioning nervous systems capable of processing pain, but pointed out that inadequately treated pain can have detrimental and permanent physiological effects. Anand emphasized the primitive biological function of pain, which is to alarm the body that something is wrong and
needs attention. Its presence from birth supports the notion that pain is often vital to survival (Hadjistavropoulos, Craig, & Fuchs-Lacelle, 2004). These reports amended attitudes towards pediatric pain assessment and management. Rather than assume children do not feel pain unless overtly indicated, researchers and clinicians inverted the paradigm to assume children always experience pain during procedures and situations that would normally be painful to an adult. As previously pointed out however, assessing pain in children (especially infants, preverbal children, and noncommunicating and/or cognitively impaired children) can be challenging, specifically when relying upon self-report. During the past several decades, emerging pediatric pain research has aimed to overcome several of these challenges.

A number of tools are now available to assess pain in typically developing children, those capable and incapable of self-report. Pediatric pain assessment tools generally rely on behavioral, physiologic, and/or facial indicators to make inferences about whether a child is hurting (Stevens, 1998). More recently developed instruments assess pain from a multidimensional perspective by including indicators from each of the aforementioned categories. When self-report is available, it is solicited. If self-report is consistent with behavioral and physiologic indicators, the most comprehensive picture of a child’s pain experience is obtained.

For groups of children where self-report is unavailable or unreliable, assessment tools must focus solely on behavioral, physiologic, and facial indicators of pain. Some of these children have traditionally been excluded from pain research—secondary to the challenges associated with assessing their pain—and include, for example, children with cognitive impairment, noncommunicating children, children with cerebral palsy, and children with other neuromuscular disorders that significantly limit their physical capabilities. Recent pain study initiatives have
resulted in a slightly increased understanding of how some children with cognitive and motor impairments experience and express pain. Several researchers have begun to describe pain expression in children with cognitive and neurological impairment, identifying through caregiver interviews and surveys those behaviors most indicative of pain (Breau, McGrath, Camfield, Rosmus, & Finley, 2000; Gilbert-MacLeod, Craig, Rocha, & Mathias, 2000; McGrath, Rosmus, Canfield, Campbell, & Hennigar, 1998; Stallard, Williams, Velleman, Lenton, & McGrath, 2002). To date however, there have been minimal attempts to comprehensively articulate how children on the autism spectrum experience and express pain. This dissertation research adds to the growing body of knowledge regarding the pain experience in vulnerable subpopulations of children; specifically, it adds to what is known about pain expression amongst the vastly diverse group of children who have been diagnosed with ASD.

**Rationale and Need for This Work**

The notable gaps in the literature indicate that while some work reflects an attempt to understand how nonverbal children and children with cognitive impairment communicate pain, the pain experience in children on the autism spectrum has hardly been explored. This is problematic because, as previously mentioned, the reported high tolerance for pain in children with ASD leads many to assume they are incapable of feeling pain. Nader, Oberlander, Chambers, and Craig (2004) published what is perhaps the first and only empirical investigation examining pain expression in children with autism. They reported that “children with autism display a significant behavioral reaction in response to a painful stimulus, and these findings are in sharp contrast to the prevailing beliefs of pain insensitivity described in the literature to date” (p. 88). Interestingly, they also highlighted the challenges parents encounter when assessing pain in their children with autism. Parents were asked to report whether they believed their children were sensitive to everyday painful events at home. When parental reports were compared to the
child’s objective facial responses to an acute pain event, those children reacting most to the painful stimulus had been previously identified by their parents as having a high tolerance for pain. This discord led the authors to highlight the need for additional exploration into how caregivers perceive their children’s ability to feel pain. Additionally, they stated that research needs to identify the factors that might influence pain expression and parental report of pain in this population. What do caregivers believe about whether their children can feel pain, and how do they assess pain in this population? Is it possible to develop a pain assessment tool specifically for use in this unique group of individuals? These were the foundational questions for this research.

Streiner and Norman (1995) point out in their introduction to health measurement scales that “most researchers tend to magnify the deficiencies of existing measures and underestimate the effort required to develop an adequate new measure” (p. 4). They subsequently state that “perhaps the most common error committed by clinical researchers is to dismiss existing scales too lightly, and embark on the development of a new instrument with an unjustifiably optimistic and naive expectation that they can do better” (p. 4). Despite the daunting and long-term nature of instrument development, a thorough review of the literature revealed that there is no existing psychometrically sound pain assessment tool available for use with children who have ASD. Relatively little is known about how this population experiences and expresses physical pain. An assessment tool that caregivers and healthcare professionals can use to determine when children with ASD are in pain is needed.

This research established a foundation for future research into how children on the autism spectrum express pain. The following discussion addresses several identified gaps in the literature, and clarifies the discordant messages circulating when it comes to whether these
children feel pain. Caregiver perceptions regarding the pain experience in children with ASD were explored in a first study, while insight from autism experts was obtained and examined in a second study. Understanding pain expression in children on the autism spectrum is important for healthcare providers—especially nurses—who often interact directly with children and families, and function frequently as advocates and educators. Ultimately, this research is important because in gaining a better understanding of how children with ASD express pain, families of newly diagnosed children can be well-informed. This information has the potential to alleviate suffering in an often silent and isolated population.

**Theoretical Framework and Nursing Metaparadigm**

Theoretical and conceptual models inform the research process by providing an organized foundation from which relational questions and hypotheses may be derived. These questions and hypotheses may be descriptive and exploratory in nature, or they may aim to test a prescribed intervention. While some theoretical models are more abstract (metaparadigms), and speak to the philosophical assumptions and/or worldview of a particular discipline, those theories utilized to guide research (middle-range theories) must provide clear definitions for the central concepts, and articulate the multiple relationships between concepts (Alligood & Tomey, 2002; Peterson, 2004). Concepts need to be linked to empirical indicators in order to provide a means for directly measuring a particular concept and its postulated relationship to other concepts (Fawcett, 2005). The research process aims to test these theoretical relationships and either support, or refute them.

**The Development of Pain Theory**

A number of theories can “house” a research program focused on the study of pain. Pain theory has evolved rapidly during the past 40 years, ignited by the introduction of a ground-breaking model by Melzack and Wall (1965), which delineated a multidimensional pain
processing system for the first time. Their model also articulated the hypothesized role of the brain (central processing) in pain, and it is now realized that the brain is a significant modulator in the pain experience. Most current pain theories emphasize the interplay between physiology, neurobiology, and psychology (cognition and emotion) in the perception and expression of pain. This research examined how children with ASD might process, perceive and ultimately express pain to others. It focused primarily on determining how to identify whether a child with ASD is in pain. This inquiry needed to be theoretically guided by a model that addresses the relationships between concepts important to processing, perceiving, expressing, observing, and measuring pain. Specifically, because of the impairments in social and communicative abilities common to children with ASD, a theory that accounts for how these capacities influence the perception and expression of pain is most appropriate for pain research in this population.

**The Communications Model of Pain**

Craig (1998) examined available pain models and concluded that current models ignored “interpersonal processes whereby the pain experience is encoded in expressive behavior, which observers must decipher [italics added] in order to appreciate the subjective nature of pain” (p. 104). He and others set forth to develop a model that would integrate various aspects of communicating pain to others, and focused specifically on how observational strategies might allow a third party to “decode” pain behaviors in an individual who could not verbally self-report pain. The resulting Communications Model of Pain (CMOP; Hadjistavropoulos & Craig, 2002) was used to guide this research.

Hadjistavropoulos and Craig (2002) depict the pain experience in an A -> B -> C model (Figure 1-1). The internal state (A, in figure 1-1) during a physically painful event is affected by intrapersonal and contextual influences. These influences include the affective and motivational components of pain, the biological nature of the brain and nervous system, and the cognitive
capacity to apply meaning and perceive pain. The experience is encoded (B, in Figure 1-1) and expressed through various voluntary and involuntary behaviors - reflecting the interpersonal component of the model. The higher mental processing pathway encompasses one’s ability to voluntarily communicate his or her pain, or to perhaps even hide the fact that one is in pain. This represents the shared experience of pain with others, particularly with an observer. Coupled with the voluntary pain expressive signals shared with others are various involuntary (or reflexive) indicators of pain. Finally, everything an observer knows about the pain experience of another individual must “come from” the individual experiencing pain. This process of decoding (C, in Figure 1-1) someone else’s experience depends heavily on the clarity of the messages coming from the verbal and nonverbal programs. Additionally, individuals bring their own experiences and perceptions with them when they attempt to decode, or interpret, another person’s pain expressive behaviors and/or reports.

The CMOP is a comprehensive model, appropriate for this inquiry into how children with ASD might experience and express pain. Its emphasis on inferring pain from verbal and nonverbal indicators, and inclusion of both voluntary and involuntary responses to pain, made this framework useful for the conducted research. Theoretical models allow researchers to test specific relationships between concepts. This requires a formal definition of each concept, and a way to empirically measure concepts. In this particular research, it was not yet determined how children with ASD might encode pain in expressive behaviors. And thus, because this research involved conducting exploratory descriptive studies, empirical indicators were not derived from the current model. Rather, this research aimed to determine what specific empirical indicators would be appropriate for each concept described by the model, given the population of interest. For example, in many children with ASD, communication is impaired, and thus children cannot
verbally self-report pain. The CMOP emphasizes the importance of identifying and utilizing automatic processing and nonverbal indicators of pain when self-report is not available. This research began to make it possible to attach specific empirical indicators (i.e., those items comprising a pain assessment tool for children with ASD) to the concept labeled “nonverbal programs” in the model.

Other concepts explored in this research include “observer bias,” “message clarity,” and for those children with verbal ability, “verbal programs” of pain expression. Caregiver perceptions regarding whether they believe their children can feel pain were solicited. These perceptions can now be examined in light of this model, and the impact of how observer bias might influence a caregiver’s ability to decode pain expression in their children can be formally tested in future studies. Additionally, “message clarity” was explored as caregivers described the challenges associated with assessing pain in their children.

The CMOP is one of the first thorough attempts to emphasize that pain processing is not comprised solely of neural and sensory components. This model highlights not only the role of the individual experience, but also the interpersonal experience. The model also allows one to consider how joint attention might influence the understanding and expression of pain in individuals. If children with ASD are capable of feeling physical pain, their intrapersonal experience might be similar to a typical child’s. It may never be possible to accurately depict how they perceive or apply meaning, because as a “decoder,” all information must be interpreted. Knowing, however, that children with ASD have difficulty jointly attending to a shared experience, one can postulate that attending to a painful event might be impaired. Dawson et al. (2004) report that attention to the distress of others is related to one’s joint attentional skills. These findings indicate that the social intersubjectivity of the pain experience
likely plays a more important role than previously considered in earlier models of pain (Deyo, Prkachin, & Mercer, 2004). If intersubjectivity is impaired in individuals with ASD, their sharing of a painful experience is also subject to impairment. The CMOP offers a creative framework which also accommodates deficits in social interaction and allows researchers to begin to theorize how social impairment might influence pain expression—which is critically important when considering the pain experience in children with ASD.

Given the challenges associated with assessing pain in children with ASD, the CMOP accommodates those modes of pain expression (involuntary and nonverbal indicators) that will likely need to be relied upon for accurate pain assessment in this population. As the exploratory, descriptive studies were conducted, findings were examined in light of how this model depicts the pain experience in individuals with communication and/or social impairments.

**King’s Interacting Systems Framework: Nursing Metaparadigm**

Parse (2000), a well known nursing theorist, emphasizes that middle-range theories are “hypothetical statements that can be tested by quantitative research through the operationalization of terms” (p. 91). She purports that these groups of hypothetical statements are without meaning until they are framed within a conceptual framework. Some nursing theorists, when addressing the advancement of nursing science, believe that middle-range theories may be “borrowed” from other disciplines only if they are linked to nursing conceptual frameworks and philosophy (Fawcett, 2005; Parse 2000; Villarruel et al., 2001). Thus, the particular pain model guiding this research was linked to a more abstract nursing metaparadigm; specifically, to the one developed by Imogene King (1981).

The general concepts and underlying worldview of the CMOP align nicely with the philosophical and conceptual development of King’s (1981) systems framework, which emphasizes interaction, and the communicative nature of human beings as they relate to others.
and their environment. Imogene King’s conceptual model defines the major concepts pertinent to the discipline of nursing, such as those in the overall metaparadigm of nursing: human beings, environment, health, and nursing. She delineates her philosophy regarding how these abstract concepts relate to one another. Her conceptual framework derives from systems theory, and focuses on three specific interacting systems: personal systems, interpersonal systems, and social systems.

The CMOP approaches pain from a framework that assumes individuals bring their entire being to a situation, and that they interact with others, conveying communicative signals (i.e., in this study, expressing pain via verbal and nonverbal modalities) for interpretation by others. The CMOP emphasizes the importance of the environment, communication, and social factors in the experience, expression, and observation/interpretation of pain, both of which are described abstractly by King (1981). The metaparadigm developed by King reflects the overall nursing approach that was applied when conducting this research. The CMOP however, more specifically defines the concepts pertinent to this research, and will ultimately allow for specific testing of relationships between concepts.

**Dissertation Purpose**

The overall purpose of this research was to explore how children with ASD express pain, and determine whether it is feasible to develop a pain assessment tool specifically for this population. In order to approach this global research question, a series of two studies were conducted to address several more specific questions.

**Research Questions**

Two exploratory descriptive studies addressed the following research questions:

1. What do caregivers of children with autism spectrum disorder (ASD) believe about the way their children experience and express pain?
2. Do caregivers find it difficult to determine when their children with ASD are in pain? Further, if they can determine when their child is in pain, what specific indicators are utilized in pain assessment?

3. Do caregivers of children with ASD believe healthcare providers adequately assess and treat their children’s pain?

4. How do autism experts describe the nature of the pain experience in children with ASD?

5. What specific indicators do autism and pain experts believe are relevant to pain assessment in children with ASD?

6. Is it feasible to develop a pain assessment tool for use in the ASD population?
Figure 1-1. The Communications Model of Pain. (Hadjistavropoulos & Craig, 2002)
CHAPTER 2
REVIEW OF THE LITERATURE

In order to appreciate the challenges encountered when attempting to determine whether a child with autism spectrum disorder (ASD) is in pain, one must be familiar with the specific nature of the disorder. The purpose of the following review is to a) provide an overview of ASD, by describing current positions regarding prevalence and proposed etiologies, and by examining the specific strengths and weaknesses associated with children on the autism spectrum, b) explore communication theory, particularly as it explains the impairments in ASD, and focus specifically on the implications of this deficit’s influence on pain expression, c) present a brief discussion of pain expression in normal children, d) review research focused on the development of pain assessment tools for use in several related vulnerable populations of children, e) explore the applicability of using these existing pain assessment tools in ASD, and f) discuss ethical issues pertinent to this research.

Autism Spectrum Disorder (ASD)

In 1943, an original paper by Leo Kanner described the presentation of children who would later be labeled as “autistic.” In his words, “There is from the start an extreme autistic aloneness that, whenever possible, disregards, ignores, shuts out anything that comes to the child from the outside” (Frith, 2003, p. 6). This aloneness emerges from a fundamental impairment in social interaction. It is this social impairment, along with impairments in communication, and the presence of restricted patterns of behaviors and interests that characterize a group of disorders known as autism spectrum disorders (ASDs; Charman & Baird, 2002; Dover & Le Couteur, 2007; National Institute of Mental Health [NIMH], 2007; Walker et al., 2004). ASDs are sometimes also referred to as pervasive developmental disorders (PDDs), and the PDDs include autistic disorder, Asperger’s disorder, childhood disintegrative disorder (CDD), Rett’s
disorder, and PDD-not otherwise specified (PDD-NOS; American Psychiatric Association [DSM-IV-TR], 2000; Volkmar & Klin, 2005). In clinical practice, Rett’s disorder and CDD are sometimes not included in the grouping of ASD, and therefore some sources may describe ASD as including autism, AD, and PDD-NOS (Kutscher, 2006). For the purpose of this review, ASD will refer to autism, AD, and PDD-NOS.

These disorders are collectively described as a spectrum of disorders because the symptoms can occur in a variety of combinations and present with varying degrees of severity (Autism Society of America [ASA], 2006). The emergence of language describing a spectrum of disorders highlights the possible interrelatedness of these diagnoses, and accommodates the nature of the variability in symptom severity often seen in children on the spectrum (Filipek et al., 1999). More recent attention to the category of PDD-NOS has contributed to the conceptualization of a “broader phenotype” of ASD, as this category includes children who display various patterns of skills and difficulties related to, but not adequately meeting the restrictive criteria for other pervasive developmental disorders such as Asperger’s disorder, or autism (Dawson et al., 2002; Filipek et al., 1999; Volkmar, Lord, Bailey, Schultz, & Klin, 2004). Despite the challenges encountered by clinicians regarding classification, all children with ASD have difficulties in the three previously described domains: social interaction, patterns of communication, and restricted and/or repetitive behaviors or interests.

Prevalence

It was assumed for many years that autism was rare, occurring at a rate of about four to five cases per 10,000 children. Prevalence reports in the late 80s and early 90s however, indicated that the rate of ASDs might be somewhere around 30–60 per 10,000 children, with about one quarter of those (10–20 per 10,000) meeting criteria for true autism (Filipek et al., 1999; Fombonne, 2003, 2005; Volkmar, Chawarska, & Klin, 2005). This alarming rise in the
reported prevalence compelled many to explore why the number of children with ASD seemed to be increasing, and sparked debate about whether there is an autism “epidemic” (Barbaresi, Katusic, Colligan, Weaver, & Jacobsen, 2005; Barclay, 2005; Fombonne, 2001; Gernsbacher, Dawson, & Goldsmith, 2005; Newschaffer, Falb, & Gurney, 2005; Williams, Higgins, Brayne, 2006). In response to the circulating concerns regarding the true prevalence of autism and related disorders, the Centers for Disease Control (CDC) conducted a study examining 8-year-old children living in 14 sites in the United States (Centers for Disease Control [CDC], 2007). Their findings state that 1 in 150 children are living with an ASD. The rate of autism in boys is about four times higher than in girls, with a reported ratio of 4.3:1 (Fombonne, 2005).

According to the CDC, using this current prevalence data, “we can estimate that if 4 million children are born in the United States every year, approximately 24,000 of these children will eventually be diagnosed with an ASD” (CDC, 2007, ¶ 2). In addition, the CDC points out that using this prevalence rate, if it has been constant over the past two decades, we can estimate that up to 500,000 individuals between the ages of birth and 21 have an ASD (CDC, 2007). After mental retardation (intellectual impairment), ASDs are the most common developmental disability (CDC, 2007). These statistics are widely adopted and currently reported throughout the United States by organizations and experts focused on autism. The prevalence of ASD is currently higher than that of spina bifida, cancer, or Down’s syndrome (Filipek et al., 1999; Muhle, Trentacoste, & Rapin, 2004; NIMH, 2007). Inadequate pain assessment in this population has national implications and potentially neglects the needs of a large population of children.

**Review of Possible Etiologies for ASD**

Unlike other medical conditions where a precise causal mechanism can usually be delineated, the etiology of ASD continues to perplex many experts, clinicians, and researchers in
the field. It is believed that autism is a neurobehavioral disorder and has no single cause (ASA, 2006; Happe, Ronald, & Plomin, 2006; Sunil, 2006). Growing evidence supports the idea that ASD may be caused by several factors, including genetic susceptibility and environmental influences. The precise nature of the interplay between genetic predisposition and environmental factors is the targeted question of many current research initiatives. The heterogeneity of the phenotype of ASD highlights the complexity scientists encounter as they examine the proposed etiologies of ASD. There is emphasis on the importance of integrating findings from a variety of disciplines attempting to understand why children with ASD present with such varying degrees of difficulty in the three primary domains. The following sections will review current research initiatives exploring possible infectious, gastrointestinal, nutritional, environmental toxin, and inflammatory, metabolic, and neuroendocrine factors and ASD. Additionally, a few psychological perspectives are briefly presented.

**Genetic predisposition**

Family studies have reported that there is a 60% concordance for classic autism in monozygotic twins versus 0% in dizygotic twins (Muhle et al., 2004; Sunil, 2006). When a broader phenotype for autism was used, the monozygotic concordance increased to 92%. These findings point to a strong genetic base for autism, but also highlight the fact that there are likely environmental or nongenetic factors influencing the expression—and thus the severity—of autism traits (Freitag, 2007; Muhle et al., 2004; Sunil, 2006; Szpir, 2006; Zafeiriou, Ververi, & Vargiami, 2007). While most experts agree there is a strong genetic component to ASD, the more controversial discussions involve the nongenetic, environmental factors that may influence the phenotypic expression of ASD. No particular environmental factors have been scientifically proven to trigger ASD, however current ongoing studies are examining the possible gene-
environment interactions that may lead to the development of ASD, and will hopefully provide more concrete evidence for particular culprits (Szpir, 2006).

**Environmental influences**

Several studies examining *parental* beliefs about the causes of autism demonstrate that there are a number of environmental factors considered to influence the development of ASD. Most commonly cited factors include immunizations (child and maternal immunization during pregnancy), environmental exposure (to infection, medications, toxins), intolerance to food (primarily those containing casein and gluten), and specific perinatal events (fetal distress or anoxia, prematurity, low birth weight, uterine bleeding or induced labor; Harrington, Patrick, Edwards, & Brand, 2006; Mercer, Creighton, Holden, & Lewis, 2006). In addition to those factors commonly mentioned by parents, other prenatal, perinatal, and postnatal factors are under investigation (Kolevzon, Gross, & Reichenberg, 2007). It is not clear how, or whether, heavy metal exposure (mercury, cadmium, lead) might contribute to ASD. It has been hypothesized that children with ASD have a diminished ability to detoxify, and therefore exposure to metals results in increased circulating levels throughout the body, and brain (Zafeiriou et al., 2007). Studies now underway are also investigating the relationship between factors such as atypical placental growth, abnormal gut tissue, inflamed tissue in the brain, maternal and paternal age, and food allergies (Kolevzon et al., 2007; Neimark, 2007). What was once considered a primary disorder of the brain is now being conceptualized by some experts as a disorder whereby the interaction between genetic predisposition and environmental factors leads to a change in cellular function. This then manifests itself throughout the body, and presents as an array of biological, neurological, and psychological abnormalities which have come to represent the autism spectrum disorders (Herbert, 2005; Neimark, 2007).
Vaccines

Issues involving vaccination continue to be a controversial topic. There are two separate, but related, concerns regarding immunizations. The first involves the MMR vaccine and the belief that administration of this vaccine causes ASD in some genetically predisposed children. In 1998, a report by Wakefield et al. described 12 children who presented with ASD and co-morbid bowel symptoms. He contended that children with regressive autism and bowel symptoms reflected a unique process linked to the MMR vaccine (Brown, Berkovic, & Scheffer, 2007; Francois et al., 2005; Silverman & Brosco, 2007; Taylor, 2006). This proposition drew enormous attention from parents and clinicians. Despite criticism regarding the study design, the retraction of this interpretation by almost every co-author of the paper, rigorous follow-up studies, and the recent reports from the Institute of Medicine (IOM) and the CDC that there is no causal relationship between the MMR vaccine and ASD, this issue remains at the forefront of many discussions and continues to be on the autism research agenda (CDC, 1999, 2000; Institute of Medicine [IOM], 2001; Murch et al., 2004; Silverman & Brosco, 2007; Taylor, 2006).

The second hotly debated concern related to vaccination involves the sheer number of vaccines children receive, and the possible relation of increased immunizations to the perceived increase in autism prevalence (Silverman & Brosco, 2007). Attention to the preservative thimerosal (which contains ethylmercury) in vaccines, has been postulated to cause neurotoxicity, and thus ASD. Removal of thimerosal, and subsequent research (IOM, 2004) does not reveal any link between immunizations and developmental disabilities, however it is acknowledged that additional research into this issue is imperative.

Psychological perspectives

During the first decades following Kanner’s description of infantile autism, there was tremendous focus on the parental role in the psychogenesis of autism. It was wrongly believed
that autism was a psychiatric disorder resulting when parents did not provide adequate love, or failed to create a supportive environment for their children (Volkmar & Klin, 2005). Early treatment therefore, involved working with children and their parents (Silverman & Brosco, 2007). This theoretical framework was challenged in the 1960’s by Bernard Rimland (1964), who more accurately alluded to the genetic component and neurological nature of the disorder (Rimland, 1964; Silverman & Brosco, 2007).

Investigation into the etiology of ASD has now crossed a myriad of disciplines including those that take a more biomedical and physiological approach. The historical roots in psychology and psychiatry however, have resulted in a long tradition of successful research into ASD. Much of what is currently realized about the cognitive nature of ASD derives from psychological literature, and many effective treatment protocols are rooted in cognitive and behavioral therapy (i.e., Applied Behavioral Analysis (ABA)). Similar to the debated issues encountered in neurobiology, developmental psychology houses a variety of theoretical positions on the etiology of the core features found in individuals on the autism spectrum. Three commonly referenced theories include Weak Central Coherence (Frith, 1989), Executive Dysfunction (Ozonoff, 1995), and Mindblindness (Baron-Cohen, 1995).

Weak central coherence. Central coherence refers to one’s ability to process information in its context; to create a higher-level meaning from a variety of sources of input (Hill & Frith, 2003). Weak central coherence would manifest itself in a display of behaviors where an individual focuses primarily on pieces of information. Several behaviors displayed in ASD support the notion that they have weak central coherence. For example, tests involving homographs (words with more than one definition, i.e., “read” or “present”) reveal that normal children pronounce words dependent on sentence context, while verbal children with ASD do not
attend to the context of the sentence (Hill & Frith, 2003). Francesca Happe (1996) has extended these sentence findings and shows that struggles with weak coherence are manifested in low-level visual tasks as well. She poses that there is a bell-shaped distribution of coherence ability within the population of individuals with ASD, and that this might account for some of the variability seen regarding this cognitive ability. The weak central coherence theory is unique in that it not only explains several of the impairments seen in individuals with ASD, but it also has explanatory power regarding the unusual strengths present in this population. The embedded figures test (where a child must pick out a small piece of a global picture) is a test where children with ASD have been shown to excel. They often perform better than typical individuals (Hill & Frith, 2003). Researchers hypothesize that a child on the spectrum is not distracted by the overall picture, and thus succeeds rather easily in these types of tasks. A number of tests and tasks have been developed to explore central coherence abilities in children with ASD. There is merit to this hypothesis in that it has the ability to explain some of the deficits and strengths witnessed in this population.

Regarding the pain process, experts have identified a number of psychological influences that may worsen, or minimize, a normal individual’s pain experience. Part of interpreting a pain stimulus involves attributing higher-level meaning to the situation. If an individual is afraid, anxious, sees blood, or anticipates something is wrong, his or her perception of the experience might worsen. If children with autism have weak central coherence, their attention to one specific perceptual process without the added contextual information, might influence what is communicated to, or decoded by, a nearby observer. In fact, it might diminish the child’s interpretation of the painful stimulus, and subsequently minimize what is communicated to, and thus interpreted by, others.
**Executive dysfunction.** Weak central coherence seems a plausible explanation for several of the behaviors displayed by individuals with ASD. It does not, however, account for all of them. Nor is there a clear link between weak central coherence and other proposed causal theories. Similarly, executive dysfunction has explanatory power for several other behaviors witnessed in individuals in this population.

Executive function refers to the ability to perform such functions as “planning, working memory, impulse control, shifting set, and the initiation and monitoring of action….,” (Hill & Frith, 2003, p. 285). The investigation of executive function in young children with ASD has historically been limited by the minimal number of tasks available to test these cognitive skills at such a young developmental age. Recent attempts have been made to increase researchers’ abilities to examine executive function in young children (Hughes, 1998). Tasks appropriate for older children, such as the Tower of Hanoi (measuring planning abilities), the detour reaching task (measuring inhibition of prepotent response), and the Wisconsin card sorting task (measuring mental flexibility) reveal that individuals with ASD display marked disabilities in these areas (Hughes, 1998; Ozonoff & Jensen, 1999). The inability to plan accurately or resist internal impulses, and the mental rigidity evident in ASD supports the postulation that impairment in executive functioning might explain the behaviors seen in this population. The severe resistance to change and repetitiveness of actions both also appear to be dysregulations in executive functioning.

**Mindblindedness.** Mindblindedness, or more commonly referred to as the lack of a theory of mind, is the theory most developed in the autism literature (Baron-Cohen, 1995; Baron-Cohen, Tager-Flusber, & Cohen, 1993). Its wide acceptance is related to mounds of empirical work supporting the main assumption that individuals with ASD lack a theory of mind. The
most significant deficits in children with ASD involve social impairment. Frequently, abnormal social interaction is the hallmark characteristic that initiates a suspicion of autism (second to abnormal social interaction is language delay). Parents notice isolated behaviors early in their child’s development. Thus, work has been done to explore the root of this social impairment, and to identify the aberrant cognitive structure responsible for the displayed behaviors.

Theory of mind refers to the ability to understand the mental states of others, and to appreciate the relationship between mental states and actions. Theory of mind has traditionally been measured by “false belief tests.” Here, the presence of a theory of mind is reflected in an individual’s ability to monitor a scenario and separate one’s own thinking from the thought process of another, when both subjects have received different information. For example, a box of crayons is presented to a child. Next, the box is opened to reveal that inside the box is, not crayons, but popcorn. The box is closed. Then the child is asked what “Buddy” (who has not seen inside the box) will think is inside. Individuals with autism commonly fail this type of task. They fail to realize that Buddy, who has not seen inside the box, cannot possibly already know that it does not contain crayons. Empirical work continues to support the exploration of theory of mind abilities in children with ASD, and yet many questions remain. The absence of a theory of mind is hypothesized to explain many of the social and communicative characteristics seen in ASD.

Because traditional theory of mind tasks evaluate the presence of theory of mind around four years old, researchers have sought to identify cognitive structures which may act as precursors to theory of mind. Uta Frith (2003) describes theory of mind using the term “mentalizing.” She poses that the “brain has an innate mechanism for processing other people’s mental states” (p. 95). If mentalizing is innate, precursors to the theory of mind measured at four
years old might be identifiable. Suggested precursors include pretend play (metarepresentation), and joint attention (reflected in eye contact and gaze monitoring) (Charman, 2003; Morgan, Maybery, & Durkin, 2003). Joint attention is posited to influence the development of a theory of mind, and also the emergence of language (Charman, 2003), which may indicate that joint attention represents a more primary cognitive structure. Its impairment results in an absent theory of mind.

Joint attention behaviors are identifiable between six and twelve months of age. They involve dyadic or triadic sharing of attention between the infant, another person, and an object or event (Charman, 2003). Joint attention abilities have been empirically associated with language and social development. While associations have been identified, less is understood about the exact neurodevelopmental mechanisms that might explain how deficits in joint attention evolve into theory of mind and language impairments. Recall also, that “early” does not always indicate cognitive structures are “innate.” Joint attention may be a direct precursor to theory of mind development, but it is not well understood how joint attention develops, whether directly out of eye contact, or by some other unidentified mechanism.

Deficits in these proposed precursors to theory of mind (joint attention and eye contact) might influence the development of appropriate pain understanding and expression in children with ASD. The following illustration adds clarity to this proposition. When a normal young child falls down—assuming that the tumble was painful for a brief moment—most often the child glances upward in search of a bystander. Once identified, the child jointly attends to the recent event (the fall). The child’s reaction is heavily influenced by the other person’s reaction to his or her fall. Fear in the face of the mother might instigate a cry. The child’s own embarrassment might result in crying. A smile on the face of the mother, or the offer of comfort
might diminish the desire to cry. Crying is an overt indicator of pain to many caregivers. The cry in this situation however, reflects an intricate system of psychological processing. The psychological processing is blunted if the child fails to jointly attend to the event of falling down. There may be no communicative interaction at all, and this may subsequently be misinterpreted that the child felt no pain during the fall. Only when it becomes unbelievable that a child could feel no pain (i.e., discovering the child has a broken limb) might a caregiver become overtly concerned. The exploration of this notion was the premise for this research, and it is now clear that there may be a relationship between the social and communication impairments in children with ASD, and their ability to express whether they are in pain.

**Interrelatedness of psychological theories.** Initially, each of these three described psychological theories originated to explain a single causal mechanism responsible for the clinical picture of ASD. However, it is becoming more apparent that no one theory can account for all phenomena seen in individuals with ASD. No single theory has successfully explained *causality*. Theory of mind approaches are limited in their ability to simultaneously explain executive dysfunction. Hughes (1998) asserts that executive functioning is crucial and primary to a child’s development of a theory of mind. Challenging this contention, is the reminder that there are other clinical subpopulations who suffer from executive dysfunction, and who do not have difficulty understanding other’s mental states (for example, schizophrenia, attention deficit hyperactivity disorder [ADHD], and frontal lobe syndrome) (Baron-Cohen & Swettenham, 1997).

Several attempts have been made to integrate weak central coherence with the absence of a theory of mind. Jarrold (2000) asserts that weak central coherence is crucial and *primary* to a child’s development of a theory of mind. Morgan, Maybery, and Durkin (2003) tested this
relationship, and conclude that weak central coherence, joint attention, and verbal ability independently discriminate children with autism. Weak central coherence alone could not predict autism group membership.

The debate remains unresolved. There are three well-established theoretical approaches in psychology, with researchers from each position invested in uncovering the causal mechanisms of autism. Newer ideas aim to incorporate the established theories, and seek to unify the different approaches. Future exploration might reveal explanations for the variability in symptom type and severity seen across the spectrum of autism disorders. In this light, it might be possible to explain why a variety of contradictory pain experiences are reported within this population.

Clinicians, parents, and experts in autism have reason to be encouraged by the vast number of research initiatives aimed at understanding the cause(s) of ASD. Understanding the various theoretical positions regarding etiology allows one to consider how pain processing might be disrupted in this population.

**Core Features Exhibited by Children with ASD**

All autism spectrum disorders are characterized by three defining features: impairments in socialization, limitations in verbal and nonverbal communication, and the presence of restricted and repetitive behaviors and/or interests (DSM-IV-TR, 2000; NIMH, 2007). Currently, general practitioners in clinical practice utilize the *Diagnostic and Statistical Manual of Mental Disorders (DSM-IV-TR, 2000)* and the 10th edition of the *International Classification of Diseases (ICD-10)* diagnostic criteria as formal guidelines for diagnosis of autism, Asperger’s disorder, and other spectrum disorders not meeting full criteria for autism. The current criteria in the *DSM-IV-TR* and the *ICD-10* classification systems allow for extreme latitude in interpretation on the part of the clinician (Volkmar & Klin, 2005).
Despite diagnostic limitations, it is important to discuss different manifestations of the core features and deficits that are associated with each of the autism spectrum disorders.

**Autism**

Typically developing infants are social beings (NIMH, 2007). Gazing at others, attuning to voices, holding fingers, and smiling are natural social behaviors in infancy. Social impairments in autism therefore, often manifest themselves in poor eye contact, and the inability to utilize nonverbal gestures to manipulate the social environment, such as pointing or sharing gaze with others (DSM-IV-TR, 2000; Dover & Le Couteur, 2007; NIMH, 2007; Volkmar & Klin, 2005). Younger children and infants do not imitate or play the same way as typically developing children. They often lack pretend play, and might show little interest in things. As they grow, children with autism often do not develop peer relationships appropriately and do not appear to respond emotionally to others. Howlin (1998) summarizes in detail the deficits seen in autism and discusses how older children lack social awareness and thus may seem isolated from the world. They appear to not have feelings, and do not always seem sensitive to others. They may also act socially inappropriate at times. For example, verbal children with autism may ask an obese woman why she is “fat.”

Communication impairments in autism manifest as either a delay in, or complete lack of, the development of spoken language. In individuals with verbal abilities, conversation skills are limited, and they may demonstrate repetitive use of language, or idiosyncratic language use (DSM-IV-TR, 2000; Volkmar & Klin, 2005). Children with autism often repeat things or ask the same question over and over again. Parents of younger children may initially report a lack of babbling, or odd speech patterns including echolalia (repeating or echoing others), and unusual tone or pitch. Many children with autism, if they do acquire some basic language abilities, have
difficulty telling others what they want or need, and this can lead to frustration, anxiety, and even tantrum-like behavior.

Restricted patterns of behavior, interests, and activities comprise the third category of impairment in children with autism. Children with autism might be preoccupied with a particular interest that is of abnormal intensity or focus. They often demonstrate repetitive play, such as in excessively lining up cars. Many children with autism have motor mannerisms, such as hand flapping, which are repetitive in nature. Change is often a stressful occurrence for them. They may have routines or patterns from which they cannot deviate.

Deficits associated with true autism permeate all aspects of the lives of the child and family members. Special consideration needs to be taken when working with these children and families, especially in healthcare settings, in order to be sensitive to the way children with autism experience the world. While the specifications for the diagnosis of autism have been delineated and widely accepted, there is controversy about how to accommodate all children who have a spectrum disorder, but do not meet the criteria for true autism. These children include those with Asperger’s disorder, and PDD-NOS.

Asperger’s disorder (AD)

The characterization of Asperger’s disorder (AD) continues to create confusion and controversy among the autism community. While related to autism and PDD-NOS diagnostically (it is included in the DSM-IV-TR and its diagnostic criteria are identical to autism, with the exclusion of the communication criteria), the “definition” of AD is argued to be unique (Filipek et al., 1999). Some have referred to AD as “autism without mental retardation,” “high functioning autism (HFA),” or “milder forms of autism marked by higher cognitive and linguistic abilities” (Klin, McPartland, & Volkmar, 2005, p. 88). Despite the DSM-IV-TR diagnostic implications that there are no language impairments in AD, it is clear that language in
children with AD disorder is not typical. Examples of current research include attempts to define whether there are distinctions between AD and HFA, and the consideration that perhaps a broader phenotype of AD might include relatively successful individuals who are not necessarily disabled (Klin et al., 2005). Children with AD share many of the same features as children with autism, but do not have a history of language delay, and usually have average or above average intellectual abilities (Klin et al., 2005; Pratt & Buckmann, 2006; Sunil, 2006).

Socially, children with AD may not appear as withdrawn as children with autism, but they tend to approach others in inappropriate or eccentric ways (Klin et al., 2005). Children with AD do befriend others, but inherent in their friendships are difficulties related to awkwardness and perceived insensitivity on the part of others. They participate in conversation, but have a tendency to only discuss topics they are interested in and fail to “banter” back and forth as in typical two-way conversation. Their inability to understand the “rules” of interaction, and poor comprehension of jokes and metaphor, can lead to feelings of isolation and embarrassment. Children with AD may have flat and emotionless speech. They are often obsessed with particular topics, may ask repetitive questions, and display concrete, literal thinking. Their circumscribed interests lead them to learn volumes of information about a particular topic in a very intense nature. For example, a child might know or want to learn absolutely everything about vacuums, from how they are built, the history of when they were invented, down to the make and model of every vacuum in existence. Some children with AD display eccentric behaviors (Belschner, 2007). Children with AD usually have a history of poor motor coordination. Parents often describe them as “clumsy.”

The intentional focus on AD and its relation to the other spectrum disorders has led to a revitalization of research examining this particular population. The heightened attention has led
to discussion about definition, causality, detailing its relationship to other spectrum disorders, and targeting interventions specifically designed to work with these children.

**Pervasive developmental disorder-not otherwise specified (PDD-NOS)**

PDD-NOS, also referred to as “atypical autism,” is referenced when there are clinically significant autistic symptoms, but not quite enough to meet the full criteria for true autism. It is not a separate disorder with different manifestations, and therefore, many of the previously described manifestations are applicable to this population. Children with PDD-NOS are sometimes considered to demonstrate a “milder” symptomatology. Diagnosing PDD-NOS is a diagnosis by exclusion, when a child cannot be categorized into one of the other autism spectrum diagnoses. One report describes children with PDD-NOS as being “midway between the autism and AD groups on IQ, measures of adaptive behavior, and language milestones” (Walker et al., 2004, p. 178). They are on the spectrum, are higher functioning than autism, have fewer repetitive behaviors than autism and AD, and have more delayed language than children with AD (Walker et al., 2004).

**Communication Impairment in ASD**

From a very young age children learn that they live in an interactive world. The world stimulates them, it responds to them, and they quickly realize how to interact with the world in order to elicit those responses from others. These basic realizations foster a sense of connectedness to the environment. To some who study the nature and development of communication, these early infant realizations are presumed to lay the foundation for the emergence of the ability to communicate. Something goes awry, however, in children with ASD, rendering them unable to communicate in the same manner as other children.

Impairments in communication are the focus of many different studies found throughout the autism literature, and have the potential to significantly alter and/or limit pain expression. If
children with autism are known to have social and communicative impairments, it is unlikely that—according to The Communications Model of Pain (Hadjistravopoulos & Craig, 2002)—their pain experience would go uninfluenced. Theoretically, these children have inherent difficulties when it comes to expressing pain. In order to thoroughly understand how the impairments seen in children with ASD might influence their understanding and expression of physical pain (bumps, bruises, burns, etc.), it is necessary to examine communication, and specifically, the development of the ability to communicate in ASD.

This section aims to describe several current theoretical perspectives found in the communication and language literature, in an effort to begin to understand how impairments in this specific domain might influence a child’s ability to express pain. Several of the long-standing debatable issues that are commonly addressed in the language development literature are presented. Theories that attempt to explain the impairments seen in children with autism are explored. Finally, a more in-depth examination of specific communication and language impairments seen in children with ASD, as they might influence pain expression, is presented.

The pain experience is a complex, multidimensional process involving different components of the brain and central nervous system. Similarly, pain expression is a complex, multidimensional process. Rather than consider how children with autism may uniquely experience pain, some individuals assume that the impairments inherent in children with ASD actually diminish or negate pain processing altogether. This is perhaps a result of the fact that many children with ASD do not express their physical discomfort in a typical manner (i.e., cry, moan, seek comfort, etc.). To date, many case studies and personal testimonies published in the literature on autism describe sensory and perceptual abnormalities (Bursch, Ingman, Vitti, Hyman, & Zeltzer, 2004; O’Neill & Jones, 1997). Some individuals with ASD do experience
alteration in auditory, tactile, and/or visual sensory processing. Caregivers describe the
difficulties they encounter when their children with autism are upset secondary to altered sensory
perception (Zeltzer & Schlank, 2005). In addition to sensory alterations, caregivers describe
unusual, or absent, responses to painful stimuli in their children with autism. Others however,
can describe unique behaviors that alert them to their child’s being in pain. Altered pain
expression is not universally observed in ASD, but most experts agree that the pain experience
appears different in this population. It is possible that impairments in communication influence
how pain is expressed in these individuals.

Overview of Communication Theory

Frank E. X. Dance (1970) reported that there are around 100 published definitions of
communication. The conceptualization of communication has been addressed from a wide
range, and diverse set, of disciplines. For these reasons, it is nearly impossible to construct one
single, comprehensive, working definition of communication. The definition must take on the
form most appropriate for the particular purpose or inquiry. Despite the diversity, many
communication experts identify and address the various properties central to most definitions of
communication. In his text, Wood (1982) describes five premises that are foundational to a
working definition of communication. He emphasizes that communication is a dynamic process,
and that it is systemic. He points out that the whole is greater than the sum of its parts, that
communication is symbolic interaction, and finally, that meaning in communication is personally
constructed. The model he believes captures these described premises is a Symbolic
Interactionist Model of Communication, which emphasizes dynamism and also the shared
experience crucial to the overall function of communication. This trend from a more action-
oriented model of communication towards an interactional, and ultimately transactional, model
of communication has been noted in other texts on communication theory as well (Budwig,
While a number of communication theorists aim to better define communication, and their work reflects the evolution and adaptation of models attempting to define communication, other theorists explore a variety of other aspects of communication. Different disciplines generate different research programs, each aimed at addressing a particular research question related to communication. Communication research encompasses all issues related to how and why humans communicate, from interpersonal communication, communication methods used in educational settings, to mass media communication. Many of these theoretical frameworks are beyond the scope of this review, as they are not directly related to the research question these studies addressed. The particular aspects of communication theory most applicable to this review include those foundations which address the social aspects of communication, and additionally, how language is used (or not used) as a means of communication. In an attempt to understand the deficits seen in children with autism, and to discuss how different impairments might influence the verbal and nonverbal expression of pain, it is necessary to examine the closely related, but somewhat separate, body of literature focused on language development and use.

**Language and Communication**

Language and communication are intimately related. The nature of the relationship however, is one of the controversial issues articulated throughout the language literature. For
example, Hoff (2005) points out that there are different camps regarding whether the desire to communicate is the motivation for acquiring language. Several theorists focus on the child’s desire to share experience and express themselves as the impetus for language emergence. Others counter argue that the complexities of grammar cannot be learned simply by the motivation to communicate, and they pose that these theorists give children too much credit when it comes to assuming they have such a sound social understanding (Hoff, 2005).

This debate is also sometimes referred to as part of the “formalism” vs. “functionalism” debate. Formalists regard language acquisition as a separate module of development, completely independent of communication (and other social or cognitive processes for that matter). This modular conceptualization derives historically from Chomsky’s articulation of language as a “mental organ,” and was further explored by Fodor (1983). Functionalists, on the other hand, tend to describe the process of language acquisition in light of the other cognitive capacities that are developing simultaneously, including the desire to socially interact and communicate. They articulate how the interaction between the use of language and the environment (other individuals), fosters the continued development of language and communicative capacity. Interestingly, some children with autism develop language skills, but fail to utilize language to successfully communicate, lending support to the notion that language and communication are perhaps in some unique way distinctly separate.

In examining the relationship between language and general cognition, Marcus and Rabagliati (2006) address the “modularity” vs. “domain-general” debate and provide insight into a possible compromise. They propose a theory whereby cognition and language share a common background from an evolutionary standpoint, and have only recently diverged due to small changes. Thus, while distinct in some ways, they are also related, and research may ultimately
shed light on how “language builds on and departs from particular aspects of domain-general
cognition” (p. 1228). Marcus and Rabagliati point out that developmental disorders such as
ASD, might be better accommodated by such an approach, and thus would explain why some
children develop linguistic ability while remaining unable to communicate effectively.

**Language Development**

When theorists move beyond examining the nature of communication and how it relates to
language, and attempt to specifically explain the development of linguistic competence in the
early years of life, several vividly different research enterprises can be identified. Despite one’s
approach towards the study of language, there are several major issues that every theory attempts
to address and which continue to be pertinent issues in the field. These issues involve the initial
state of knowledge regarding language, the process of developmental change, and the role of
external input influencing the development of language. The follow paragraphs highlight the
most debatable issues that have historically laid the foundation for different “camps” within
language development theory and research.

**Innateness**

One of the most long-standing debates involves the issue of innateness. Rapid
development regarding the study of children’s language followed a publication in the 1950s by
Noam Chomsky. The debate that ensued between Chomsky (a nativist) and B. F. Skinner (a
learning theorist) facilitated the emergence of two divergent positions regarding the origins of
language. Chomsky focused on the grammar of children, and posited that there is an innate
Universal Grammar (UG) whereby interaction with this UG (input from environment), and the
presence of innate constraints, allows for the growth of a knowledge of language. Nativists point
to the ease and rapidity with which children acquire language, and also argue that even without
implicit instruction children develop language abilities (Meltzoff, 1999). Skinner however,
espoused that the environment shapes the development and emergence of language without the original presence of anything innate. The current state of this framework is usually referred to as connectionism. This initial debate ignited and fueled language research, bringing it ultimately to the body of knowledge that is explored and expanded today.

Current theoretical positions have, in a sense, rejected the pure behaviorist approach. Almost everyone agrees that there is something innate about the acquisition of language, and that it is the interaction of the innate capabilities with the environment that best describe its emergence. The debate currently ensues regarding the nature of the innateness and the interaction, and it is in this realm that most theories currently differ.

**Developmental changes**

In order to understand what is *innate* in language, theorists must describe the process by which infants and children take that innate capability and develop language. Explanations for the process by which individuals learn language vary significantly. Several particular issues are commonly addressed when referring to the development of language. These include explaining the process by which children acquire language, considering whether there are particular “critical periods” during the development of language, and examining the influence of “input” from the world on language development.

**Processes**

A variety of models attempt to detail the mechanism by which language abilities emerge. Modern research programs utilize newer technology and methodology and have been able to gain detailed insight into the processes by which children learn language. One segment of theoretical work focuses on babies’ abilities to identify patterns, learn rules, and even utilize statistics during language development. Work by Saffran, Aslin, and Newport (1996) emphasizes the unique ability of babies to identify patterns and use a form of statistics to
organize the sounds they hear and develop language. Pinker has written—in light of learning rules and setting parameters—that babies however “are not acquiring dozens or hundreds of rules; they are just setting a few mental switches” (Akhtar, 1999, p. 340). Pinker also focuses on how children link rules based on an innate knowledge of word categories. Patricia Kuhl (2000) accounts for all concepts: rules, patterns, and statistics; in addition she identifies other influences, such as experience, that likely shape language development. She concludes that “they use inherent learning strategies that were not expected, ones thought to be too complex and difficult for infants to use” (p. 11856).

Recently, Kuhl examined speech and linguistic processing in children with autism (Kuhl, 2005). Conclusions from this work indicate that there is a link between social interaction and language learning. This notion that social interaction is primal to the development of communication and language is historically based in writings by Vygotsky (Budwig et al., 2000). The process by which social functioning influences the development of communication and language is theorized by several different well-known researchers (Bruner, 1983; Snow, 1999; Tomasello, 1992). These frameworks are often referred to as social-pragmatic approaches. Tomasello’s approach however, has been referred to as cognitive-functional linguistics, and most recently has also been called usage-based linguistics (Tomasello, 2003). The details of his theory will be discussed later in light of explaining particular features of autism.

Critical periods and continuity

One issue commonly addressed by communication and language theorists involves the conjecture that there are critical periods in language development, where children are able to acquire and use language rapidly. Beyond these critical periods, it is proposed that language processing is different. The critical period hypothesis is supported by several different arguments. One argument highlights the fact that children learn language at an extremely rapid
pace, and also points out that children are able to acquire a second-language with greater ease than adults. Additionally, isolated case reports of children raised without exposure to language, highlight how the children remain mute and acquire relatively little language ability if intervention occurs after a certain point in development.

Those who disagree with the conceptualization of a critical period emphasize the continuous nature of communication and language development, and present counter arguments to the points mentioned above. They discuss how adults learning second languages have a longer history of utilizing and working with their first language, which interferes with the ability to learn the second language. They also point out that children in general, across a variety of skills and cognitive capacities are more flexible learners, aiding their ability to acquire language faster than adults. Patricia Kuhl (2000) highlights the learning strategies she has found employed by infants and “reinterprets the critical period for language” (p. 11850), stating that the “critical period for language depends on experience, not just time” (p. 11855). By experience, she refers to the neural commitment that is a result of experience.

Each theoretical perspective on communication and language theory generally accounts for the previously discussed issues: the nature of what is innate, and how the innateness translates—via different learning processes and mechanisms—into a functional language used to communicate. Additionally, in describing proposed processes and mechanisms, theorists address historically debated issues that have divided communication and language researchers such as continuity vs. discontinuity, and the critical period hypothesis. The following section will explore how theories that emphasize the social-cognitive nature of communication and language development explain the deficits seen in children with autism.
Communication Impairment and Its Influence on Pain Expression in ASD

Social and communicative deficits are core features of autism spectrum disorders. Many theorists attempt to explain the unique features seen in these children in light of the theoretical frameworks previously described. Those interested primarily in communicative impairments will re-visit what is believed about normal patterns of language development, and speculate where deviations in autism occur. Despite various theoretical perspectives on language origin and development, there are several widely accepted beliefs about communication and language in ASD. As theories are explained in greater detail however, differences frequently emerge.

It is generally agreed that an initial impairment most likely occurs early in development (Lord & Paul, 1997). The impairment must lend to a range of consequences, because in autism, language abilities are highly variable. Language impairment is related to, but not necessarily a result of, social and cognitive function (Lord & Paul, 1997). Despite deficits, progress within each domain of language usually follows a similar pathway as that of typically developing children. Lord and Paul (1997) refer to two general orientations towards autism: those with the modular approach (theory of mind, central coherence, and metarepresentation frameworks), and those with an information processing approach (executive function). They resolve that both orientations address how communication development is altered, and do so differently. Martin and McDonald (2003) address pragmatic language disorder and explore how it relates to weak coherence, theory of mind, and executive dysfunction. They conclude that all three processes can predict pragmatic language disorder, but do not specifically address the underlying mechanisms responsible for the communication impairment.

In examining the course of communicative competence and language development, theorists attempt to identify early indicators of communicative impairment, and have identified preverbal behaviors that are directly related to the subsequent development of more complex
communicative, and eventual linguistic abilities. Mitchell et al. (2006) found that communication and language delays can be predicted by monitoring for delays in specific gestures. Other preverbal and social abilities strongly linked to the development of language and other communicative abilities include joint attention skills. Examples of joint attentional skills that children with autism either lack, or have difficulty with, include referential looking, declarative pointing and showing, looking where others point and look, and social referencing (Carpenter & Tomasello, 2000). Joint attention is a precursor for other capabilities, such as engaging in role-reversal imitation, and understanding communicative intentions.

Tomasello (2003) published a usage-based theory of language acquisition and thus produced one of the most comprehensive theories of communication from the functionalist perspective. He focuses on defining how one understands the communicative intentions of others, and desires to share or direct the attentional states of others. This, according to him, facilitates the emergence of language. He emphasizes the communicative nature of interactions, even if there is minimal language exchange, and focuses on how the interaction itself can teach additional communicative skills. He also links gestures, such as pointing, to the emergence of an intersubjective experience from which linguistic symbols are used to communicate.

These basic, foundational social-cognitive skills are altered in individuals with ASD. What remains unclear is exactly how, or to what extent, these abilities are impaired. One of the most perplexing concerns to those who work in the field of autism is the extreme variability found along the spectrum, including variability in the communicative and language abilities of individuals. Most theories are unable to completely address this particular issue. As a result, it is difficult to generalize research findings. Interventions and treatments therefore, must be tailored to each specific child. Additionally, the variability implies that each child’s pain
experience, and thus ability to express his or her pain, may be a highly individualized phenomenon. This review of communication impairments in autism explores how particular deficits in social-cognitive and communicative skills might relate to the pain experience, and allows one to begin to approach the pain experience in children with ASD thoughtfully.

**Pain Assessment in Children**

The body of literature regarding the pain experience in typical, healthy children has grown rapidly over the past two decades. Significant advances have improved the assessment, measurement, and treatment of pain in children of all ages. National organizations dedicated to the care of children have begun to acknowledge the prevalence of pain (acute, chronic, procedural, etc.) in this population. International efforts to examine the prevalence of pain in children, and attempts to characterize their pain have allowed investigators to begin to quantify the presence of pain in children and adolescents. A recent article by Roth-Isigkeit, Thyen, Stoven, Schwarzenberger, & Schmucker (2005) from Germany published pain prevalence rates as high as 83% (n=739) when children were asked to report if they had experienced pain in the previous three months. Of children in the United States, 20% (aged 5-17) suffer chronic headaches (Zeltzer & Schlank, 2005). Zeltzer & Schlank (2005), experts in managing chronic pain in children, also report that juvenile arthritis is one of the most common chronic diseases in childhood, and that the prevalence of fibromyalgia is at least as high as 6% in school-age children. It is estimated that up to 20% of children suffer from some sort of chronic pain condition (Bassett, 2005). As the understanding of the prevalence and nature of pain in typically developing children increases, additional work into assessing and managing pain in subpopulations of vulnerable children becomes imperative.

Currently, there is ample research devoted to developing, validating, and modifying tools for assessment and measurement of pain across all ages in typically developing children. There
are behavioral- and physiologically-based tools for neonates and infants (Johnston, Stevens, Boyer, & Porter, 2003), and there are now developmentally appropriate self-report tools that have been modified for young and school-age children (Champion, Goodenough, von Baeyer, & Thomas, 1998; Gaffney, McGrath, & Dick, 2003). These instruments have been adapted and validated for use in a variety of settings (i.e., inpatient, outpatient, school) and under a variety of circumstances (i.e., children with cancer, or children after surgery). There are tools available to characterize the quality of different types of pain (i.e., acute, chronic, neuropathic), and also to monitor the effectiveness of different treatments (i.e., measuring for changes in pain scores). Additional bodies of literature focus on chronic pain management in children, while some even focus on the pain associated with specific disease processes. The national recognition of the importance of pain assessment and management in children has led to the establishment of pain clinics, where entire teams of professionals are dedicated to helping children in pain.

It is unreasonable to think that children with disabilities or developmental disorders are “exempt” from the pain prevalence reports described earlier. Alternatively, several pain experts posit that the prevalence rates of pain would logically be higher in these children (McGrath, 1998). Stallard, Williams, Lenton, & Velleman (2001) report that everyday pain in cognitively impaired, noncommunicating children is common, yet rarely treated. When specifically considering children on the autism spectrum, one needs to recall the commonly reported hypersensitivities and the variety of behavioral tendencies these children exhibit. Zeltzer & Schlank (2005) propose that children with already altered neurological systems will experience, and thus respond to, basic pain sensations differently. Bursch et al. (2004) posit that abnormal sensory processing will not eliminate the ability to feel pain, but it might affect the cognitive interpretation of the sensory experience. Additionally, in children with ASD, several
investigators propose that the anxiety often experienced by this population might actually *contribute* to the presence of pain, especially in those whose muscles remain tense for long periods of time (Zeltzer & Schlank, 2005).

Many children in these vulnerable populations endure medical procedures and everyday painful events similar to typically developing children, if not in greater frequency. It is imperative that the current movement to examine and better understand pain expression and management in these groups continue. In preparation for discussing the development of an instrument for use in ASD, the remainder of this literature review closely examines the research that informs what is known about pain assessment in some similar, but distinctly different, populations of children—those with cognitive, neuromuscular, communication, and/or other developmental disabilities.

**Pain Assessment in Vulnerable Pediatric Populations**

Despite the wealth of information available on pediatric pain assessment, there is a paucity of research focused on pain assessment in children with cognitive, neuromuscular, communication, and/or developmental disabilities. While children with ASD display some similar characteristics, and it might appear on the surface that a single pain assessment tool would be appropriate for all of these subpopulations of children, there are significant differences warranting an investigation of whether children with ASD require a distinct pain assessment tool. Children with ASD are not always cognitively impaired, and the combination of their social and communicative impairment is unique to this population.

Initial studies examining pain expression in children with disabilities reveal that it is inappropriate to use pain assessment tools currently available for typically developing children with these populations (Oberlander & Craig, 2003). Children with disabilities commonly exhibit cognitive, motor, neurological, and communicative impairments which confound traditional
assessment tools and lend to inaccurate measurement of pain. For example, facial and spastic movements, guarding, groans, and tears may be associated with pain expression in the typical, healthy child. Some children with disabilities however, display these behaviors as part of their daily, nonpain repertoire of expressions (Oberlander & Craig, 2003). It is therefore impractical to consider these behaviors as indicative of pain in all children with developmental disabilities (Breau et al., 2000). Regarding self-report measures of pain, many children with cognitive and communicative impairments are unable to look at a series of faces (i.e., the Faces Scale) and self-report which face reflects how they feel at the time.

In the mid-90s, after realizing the limitations of available pediatric pain assessment tools, several researchers focused on developing assessment tools applicable to specific groups of children; namely, those children with various developmental disabilities. Behavioral assessment tools have subsequently emerged in the literature, and are now available for use in children with a) cerebral palsy (CP), with or without severe neurological impairment, b) infants and neonates with—or at risk for—neurological impairment, c) children with cognitive impairment and/or developmental delay, and d) children with cognitive impairment and the inability to communicate verbally. While these populations are somewhat similar in nature, and researchers acknowledge overlap, they also emphasize the importance of continuing to validate pain assessment tools in each specific population. The following sections review pain assessment tools currently available for these vulnerable populations of children.

**Cerebral Palsy and Neurological Impairment**

Giusiano, Jimeno, Collignon, and Chau (1995) first examined pain expressive behaviors in children with CP and neurological impairment. They developed a pain assessment tool comprised of 22 items and called it the Evaluation Scale for Pain in CP. They derived their items from behaviors that physicians identified as indicative of pain in children with CP during a
physical exam. The initial publication of this scale did not provide information on its psychometric properties. A follow up study by Collignon and Giusiano (2001) however, indicates that they have further developed and tested the scale, creating a 10-item list of pain expressive behaviors for use in children with CP. They discuss validation of this newest version of the scale, and report that it demonstrates good psychometric properties. The ten identified pain indicators include: “crying during manipulation,” “painful expression,” “interest in the surrounding,” “increased tonus problems,” “coordinated defensive reaction,” moaning or inaudible cries,” “inability to communicate with nurse,” “increase in spontaneous movement,” “protection of painful areas,” and “observing pain-easing posturing.” The final ten items reported by this group are similar to findings by McGrath, Rosmus, Canfield, Campbell, and Hennigar (1998). These researchers were developing a pain assessment scale at the same time. Their scale, and its subsequent development, is discussed in a later section.

Hadden & von Baeyer (2002) extended the findings of Giusiano et al. (1995) and McGrath et al. (1998) and worked to further explore the previously identified pain indicators in children with CP—specifically looking at those children with CP and severe neurological impairment. They sent surveys (The Caregiver Pain Survey) to parents of children with CP and asked 27 probing questions about pain expressive behaviors in their children. They incorporated 24 of the indicators included in the assessment tool developed by McGrath et al. (1998). The most frequently reported pain indicators include: “crying,” “less active,” “seeks comfort,” “moaning,” “not cooperating, irritable,” “stiff/spastic/tense/rigid,” “decrease in sleep,” “difficult to satisfy or pacify,” “flinches or moves body part away,” and “agitated/fidgety.”

**Neonates at Risk for Neurological Impairment**

Several publications have emerged from a group of pain researchers describing the pain experience in neonates at risk for severe neurological impairment. Stevens et al. (2006) describe
a process whereby 14 pediatric pain experts determined the usefulness of 39 pain indicators used to assess pain in the neonatal population. The original list of 39 indicators was developed by the investigators. After two Delphi rounds with experts, they identified seven indicators that had high ratings of usefulness and importance. These include: “brow bulge,” “facial grimace,” “eye squeeze,” “inconsolability,” “increased and/or fluctuations in heart rate (from baseline),” and “reduced oxygen saturation.” These identified behaviors support what has already been determined about pain assessment in neonates—that assessment relies heavily on facial and physiologic indicators. Their study reveals that even with neurological impairment, or other neurodevelopmental disorder diagnoses, pain assessment involves observing for specific facial responses and monitoring for changes in physiologic parameters.

**Children with Cognitive Impairment**

Not all children with profound cognitive impairment have simultaneous motor difficulties (like those seen in CP). Children with cognitive impairment—whose daily functioning is typical in regards to movement and activity—comprise another population where preliminary pain assessment work has been conducted. Terstegen, Koot, de Boer, & Tibboel (2003) developed an item pool of 138 potential pain indicators for this population (based on a quasi-qualitative study). While all children had profound cognitive impairment, not all children suffered from motor impairment. They subsequently observed (by video) children post-surgically, to watch for how often the potential pain indicators were present. Twenty-three reliable, observable indicators showed sensitivity to pre and post surgical observations. The most sensitive indicators were from the category “Facial Expressions,” which includes 10 behaviors. Other categories include “Motor Behaviors” (i.e., “tensed up”), “Social Behavior/Mood” (i.e., “panics,” “accepts comfort”, and “rebellious”), “Vocalizations,” and “Physiological Signs.” For the first time, Terstegen et al. (2003) speak to the social-communicative impairments sometimes seen in
children with cognitive impairment, and thus first identify another dimension important to consider in pain assessment.

**Non-Communicating Children with Cognitive Impairment**

The development, and subsequent publication, of these various pain assessment tools has been somewhat simultaneous. Thus, there is overlap within the research, and it is difficult to describe how each assessment tool builds upon the others. The benefit however, of having tools emerge around the same time for similar populations, is that comparison of each instrument reveals stark similarities and differences. There are solid commonalities amongst the tools available for each population, but there are also subtle differences that speak to the nature of each specific population. This premise supports the argument that a pain assessment tool specifically for use in ASD is warranted.

One of the more widely available, well developed and validated instruments available for use in children with cognitive impairment and communication impairment is the previously mentioned assessment tool developed by McGrath et al. (1998). McGrath and colleagues utilized caregivers of children with CP to generate a list of behaviors they typically use to discern the presence of pain in their children. Of note, none of the caregivers interviewed had a child with ASD. They identified 30 behaviors and put them into 7 categories, which include “Vocal,” “Social,” “Facial,” “Activity,” “Body and Limbs,” “Physiological,” and “Eating/Sleeping.” In a subsequent study, Breau et al. (2000) further tested this checklist, now titled the Non-Communicating Children’s Pain Checklist, utilizing a group of 32 caregivers. Eight of these caregivers had a child with “autism/PDD,” however no children with ASD are reported to have been included in any of the subsequent validation studies (Breau, Finley, McGrath, & Camfield, 2002; Breau, McGrath, Camfield, & Finley, 2002). Preliminary validation indicates that the checklist exhibits internal consistency, and can detect pain and is reliable over time. They note
that the checklist needs further evaluation, specifically in other groups of children with impairments, and that it should be determined whether the checklist would be helpful to providers in the clinic or inpatient setting who might not know the children intimately.

A revised version, The Non-Communicating Children’s Pain Checklist – Revised, (Breau, McGrath, Camfield, & Finley, 2002) and a postoperative version, The Non-Communicating Children’s Pain Checklist – Postoperative Version (Breau, Finley, McGrath, & Camfield, 2002) have been subsequently validated and are now available. The revision did not change any of the thirty pain expressive behaviors, but simply re-worded several of the items, and changed the response format for each item from present/absent, to a 4-point Likert scale. The postoperative version is similar to the original checklist; however it does not contain the “Eating/Sleeping” items because these are not directly relevant to postoperative pain in the clinical setting. The postoperative version demonstrates good psychometric properties and it is reported that this scale is valuable when assessing pain in children with intellectual impairments postoperatively.

Additional development from this initial work with the Non-Communicating Children’s Pain Checklist has resulted in a) identification of a subset of seven items (“not cooperating, cranky, irritable, unhappy,” “seeks comfort or physical closeness,” “change in eyes, including squinching, eyes wide open, eye frown,” “not moving, less active, quiet,” “gestures to or touches part of the body that hurts,” “tears,” and “gasp”) that can successfully predict pain in children with cognitive impairment who cannot communicate (Breau, Camfield, McGrath, Rosmus, & Finley, 2001), and b) another attempt to develop a checklist aimed at assessing chronic pain in children without CP—but who have a wider range of cognitive impairment and the inability to communicate (Stallard, Williams, Velleman, Lenton, & McGrath, 2002).
This development by Stallard et al. (2002) involved caregiver interviews whereby 203 pain cues were identified and subsequently examined by experts. They identified six cues used by 90% of caregivers as indicators of pain. These include: “crying,” “screaming/yelling,” “distressed face,” “tense body,” “difficult to comfort,” and “flinches if touched.” The investigators report that despite asking about chronic pain, these cues tend to identify acute pain responses, and they highlight the difficulties encountered when assessing chronic pain in these populations. They also mention that even though this scale was developed utilizing parents of children with a wider range of cognitive and communicative impairments, its usefulness in some groups of children—specifically children with ASD—is still questionable and needs to be explored.

Stallard et al. continued work with these six identified cues and tested the instrument they developed and titled the Pain Indicator for Communicatively Impaired Children (PICIC). They used 67 caregivers of children with central nervous system disorders (no children with ASD). They found that 5 cues were correlated with caregiver judgments of pain, and that the presence of one observable cue (“screwed up or distressed looking face”) correctly identified almost 72% of the children in pain, and 93% of the children not in pain. Interestingly, crying was not a useful indicator of pain, given that many parents acknowledged the presence of crying in a number of distressing situations other than pain. One of the more novel findings in this particular work was the addition of pain cues by caregivers. Caregivers noted that he or she sometimes “pushes me,” “bites bottom lip,” “moves from one place to another,” “claps his hands,” “bangs head,” “laughs,” “rubs head,” and/or “bites,” when in pain. The PICIC might be a useful caregiver tool, but may not be helpful to individuals unfamiliar with the child, and the additional items noted by caregivers speak to a) the intimate knowledge caregivers have
regarding pain in their children, and b) the fact that pain expression in children with cognitive and communicative impairment may be quite individualized.

Acknowledging the individualized nature of pain expression in these special populations, Solodiuk and Curley (2003) developed The Individualized Numeric Rating Scale (INRS) to help assess pain in children with cognitive impairment and communication difficulties. Their assessment scale is not a checklist, but rather resembles a process by which a child’s personal pain profile might be generated. It asks caregivers to think about their child’s past painful events, provides specific indicators they might take note of regarding facial expression, body movements, activity, social interaction, cry, and consolability, and then asks caregivers to describe what their child does when in: no pain, moderate pain, and severe pain. The resulting “picture” of the child in pain is then provided to individuals involved in his or her care (i.e., nurses, physicians, teachers, therapists, etc.). This novel approach speaks to the uniqueness of each child’s individual expression of pain, however it requires that a caregiver know what his or her child does when in pain.

Finally, two additional qualitative studies directly address the experience of pain in children with special needs and severe neurological impairment (Carter, McArthur, & Cunliffe, 2002; Hunt, Mastroymannopoulou, Goldman, & Seers, 2003). These qualitative studies report that there are three forms of knowledge required for optimal pain assessment and management: knowing the child, knowing the population, and knowing the science (Hunt et al., 2002). Carter et al. (2002) highlight issues important to caregivers of children with special needs regarding pain management. They report four emerging themes: “learning to live with pain,” “dealing with uncertainty,” “expression of pain,” and “making decisions.” None of the parents had been taught
about pain management in their children; they had to “learn.” Parents discussed the lack of information available and reinforced the need for individualized pain assessment tools.

Information regarding the pain experience in a few other populations is available, but scarce, and does not involve development of pain assessment tools. For example, there are reports of children with Down’s syndrome having delayed processing of pain and a latent behavioral response to painful stimulus…but there is no pain assessment tool specifically developed for use in children with Down’s syndrome (Hennequin, Morin, & Feine, 2000). As previously discussed in Chapter 1, there is one study indicating that children with ASD have a significant facial response to painful stimuli, but their means of assessing the acute pain event involved utilizing an intricate and detailed facial coding scheme—one that is impractical to use on a regular basis for pain assessment in ASD (Nader et al., 2004).

Challenges to assessing pain in children with ASD involve the sometimes held perception that children with ASD cannot feel pain, and thus this research aimed to determine whether caregivers could assess pain in their children with ASD. From there, it can be determined what type of pain assessment tool would best benefit this population. This review of available pain assessment tools, and the examination of research looking at pain assessment in vulnerable populations of children, highlights the importance of continued pain assessment research in these unique populations. It is apparent that pain expression in children with cognitive impairment, communication difficulties, developmental and neuromuscular disorders is somewhat variable. Several core pain expressive behaviors have been identified for these populations, however additional work is required to ensure that pain assessment is valid in each of the different groups of children. While children with ASD might exhibit several of the already identified pain expressive behaviors, close examination of this population is warranted. The available literature
provides a foundation from which additional research into pain assessment in children with ASD may stem.

**Ethical Considerations**

This next discussion illuminates several ethical issues specifically relevant to the conducted research. It briefly presents the ethical principles pertinent to these issues, and describes measures taken to minimize risk to study participants. The first issue relates to the fact that this research involved vulnerable populations (children, children with developmental disabilities, and caregivers of children with developmental disabilities). The second issue relates to the nature of the research question—which involved the study of pain.

**Vulnerable Populations**

Autonomy refers to recognizing an individual’s “capacities and perspective, including his or her right to hold certain views, to make certain choices, and to take certain actions based on personal values and beliefs” (Beauchamp & Walters, 1999, p. 19). In an effort to preserve a participant’s autonomy, most research involving human subjects requires that the participants provide informed consent. The ability to give informed consent requires that a) participants be given all information regarding their participation, b) participants realize their participation is voluntary, c) participants understand all the information given to them, and d) participants have the capacity for decision-making (Ascension Health, 2007). Special considerations were made to ensure these requirements in the conducted studies. While the first stages of this research involved primarily the caregivers of children with ASD, future studies might involve more active participation by the children themselves, and thus issues pertinent to these future studies are also reviewed in this section.

Whenever possible, children need to be included in the informed consent process (American Academy of Pediatrics [AAP], 1995; Helseth & Slettebo, 2004). Legally, minors
cannot enter into a contract, and thus they require parental proxy. Ideally, even though minors cannot give consent, it is useful to obtain assent. Assent is the “moral requirement to acquire the closest approximation of consent one can achieve within the child’s capacity to understand” (Helseth & Slettebo, 2004, p. 302). Research involving children with ASD poses an additional challenge to assent when one considers the limited communication abilities of the population. Because ASD is a heterogeneous disorder, some children may be more capable of understanding their role in participation than others. It is important to work with parents to ensure that their proxy consent is informed, and that if it is possible to obtain assent, this is done. The first study conducted in this research utilized surveys, which were filled out solely by caregivers. An informational letter was presented with the survey, describing the voluntary nature of filling out the form and returning it. According to the Institutional Review Board (IRB) at the University of Florida, consent may be implied if the caregiver fills out the form and then voluntarily sends it back. The requirement for documentation of informed consent may be waived (University of Florida Institutional Review Board, 2003/2004), and was approved for this research.

Working with caregivers of children with ASD was also a unique situation. Parents of children with developmental disabilities need to know that their participation in research is completely voluntary, and that the care of their child in no way depends on participation in studies. If there is any sense that they must participate in order to receive quality care for their child, ethical principles involving justice may be violated. There are several principles of justice which are typically contextualized when discussed (Beauchamp & Walters, 2003). Beauchamp and Walters (1999) point out the minimal, beginning principle as being concerned with ensuring that “like cases should be treated alike, or, to use the language of equality, equals ought to be treated equally and unequals unequally” (p. 22). Research with parents of vulnerable children
requires that when recruited, these parents are ensured that the care of their child does not change with or without their participation. Utilization of institutional review boards can help the researcher examine the process by which participants (parents and their children) are protected.

The Study of Pain

When working in fields involving the clinical treatment and research of pain, there is an ethical impetus to relieve pain. Hadjistavropoulos (2004) points out that regardless of one’s ethical perspective, ensuring that vulnerable populations have access to adequate pain assessment and management is *not* usually a controversial issue. Walco, Burns, and Cassidy (2003) point out that numerous papers assert that the alleviation of pain is an ethical imperative in nursing and medicine. According to Ferrell and colleagues (2001), the undertreatment of pain in vulnerable populations (children, cognitively impaired individuals, elderly), is a top ethical concern among physicians and psychologists who manage pain. The ethical principle of beneficence, and its relationship to nonmaleficence, is pertinent to examining the phenomenon of pain. According to Beauchamp and Walters (2003), beneficence is a complex principle, involving one’s obligation to 1) actively promote good, 2) prevent evil or harm, 3) remove evil or harm and 4) do no harm (nonmaleficence).

In an attempt to ensure that children with communicative and social impairments are not experiencing unrelieved pain, this research could be considered an ethical imperative. The studies conducted did not involve purposefully inflicting pain upon participants, and because of the vulnerability of this population, this would never be an acceptable research method. Any examination of children in pain would have to occur in a natural setting, whereby the child might be in pain from some everyday occurrence, or perhaps because of a necessary medical procedure. Similar to pain research in the elderly with dementia, prescribed protocols ensuring that no unnecessary harm is inflicted on participants will need to be developed. Effective forms of pain
relief must be provided if pain is detected at any time (Hadjistavropoulos, 2004). There are
current ethical codes available to guide research in the domain of pain, published by a variety of
regulatory organizations, and several organized groups focused on the study of pain—the
American Pain Society [APS], for example. This research was guided by ethical codes, and all
research involving parents and children underwent review by the IRB, whose sole purpose is to
protect research participants.

**Rationale and Need for Instrument Development**

The literature does not report the existence of a valid, reliable instrument to assess for—and
subsequently measure and manage—pain in children on the autism spectrum. Any attempt
to utilize an existing tool, or adapt a tool for this population is confounded by the considerable
confusion surrounding whether children with autism are even capable of feeling pain. It is not
known what specific parts of the pain process might be impaired or influenced by their inherent
limitations. Because of the confusion that exists regarding how children with ASD experience
pain, it was inappropriate to attempt to design an assessment tool without a better understanding
of the phenomenon at hand. The studies conducted in this research were necessary in order to
determine whether it is appropriate to continue instrument development. They a) explored
caregiver perspectives regarding pain expression in children with ASD, b) generated a number of
potential pain expressive behaviors appropriate for an initial item pool, and c) reduced the initial
list of behaviors to a subset of items that can now be used to further develop a tool for assessing
pain in this population.

**Summary**

A review of autism (its prevalence, theoretical positions regarding etiology, and
description of core features associated with the disorder) prepares the reader to consider how
pain expression might be different in this population. Additionally, a focused theoretical
discussion on the nature of communication, communication development, and the
communication impairments specific to autism allows one to realize that self-report of pain is
limited in this population. Detailed examination of work that has been done to examine pain
expression in similar and related populations of children provides an understanding of the
foundation from which this current research extends. Finally, discussion of the ethical
implications related to a) working with vulnerable populations, and b) the study of pain, ensures
the reader that this Investigator minimized risk to study participants when conducting this
research.

This review describes pain assessment tools available for children with cognitive
impairment, children with neuromuscular disorders (i.e., CP), and children with communication
impairment. These assessment tools provide valuable insight into how families and healthcare
providers assess and evaluate pain in unique children. None of the aforementioned instruments
however, focus solely on children with autism spectrum disorder. While children with ASD do
have limitations in communication, the breadth of their limitations extends beyond
communication, to include the social and behavioral impairments previously described in detail.

It is inappropriate to use existing instruments to assess pain in this population.
Furthermore, several of the indicators included in the more recently developed instruments for
children with communication impairment may not be applicable to children with autism (i.e.,
“seeking comfort or physical closeness”). If one watches for these specific indicators, and does
not see them, it may be wrongly assumed that a child with ASD is pain free. This review
highlights the theoretical uniqueness of the pain experience in ASD, and supports the premise of
this research: that it was necessary to develop a pain assessment tool specifically for use in this
population.
CHAPTER 3
METHODS AND PROCEDURES

The purpose of this research was to explore how children with autism spectrum disorder (ASD) express pain, and to begin development of a pain assessment tool for use with this population. A series of two studies were conducted to a) describe how caregivers of children with ASD determine when their children are in pain, b) understand what experts believe about pain expressive behaviors in children with ASD, and c) establish the content validity of a set of pain expressive indicators that may be used to assess pain in children with ASD.

Instrument Development

DeVellis (2003) articulates eight steps necessary to develop a psychometrically sound instrument. They include a) determine what is to be measured, b) generate an item pool, c) determine the format for measurement, d) have the initial pool reviewed by experts, e) consider the inclusion of validation items, f) administer items to a development sample, g) evaluate the items, and h) optimize the scale length. Measurement experts (Allen & Yen, 1979; Crocker & Algina, 1986; Portney & Watkins, 2000) emphasize the extreme importance of the initial steps in instrument development. This research aimed to complete the first four steps in developing a pain assessment tool for use in ASD. These steps are critical for future research, which will continue the development and testing of a pain assessment tool for use in this population. Thoughtfulness early in the development process ensures that an optimally valid instrument results.

Study 1: Caregiver Perspectives of the Pain Experience in ASD

The first study conducted for this research was titled “Understanding the Pain Experience in Children with Autism: Caregiver Perspectives,” and asked caregivers to provide information
about how they determine when their children with ASD are in pain. From this point on, this first study will be referred to as “Study 1” and the second study will be referred to as “Study 2.”

**Purpose**

The purpose of this exploratory descriptive study was to characterize caregiver perspectives and concerns regarding pain in their children with autism, and to begin to identify pain expressive indicators and behaviors unique to children with ASD. This was achieved by examining caregiver responses to a mailed survey and by conducting semi-structured interviews with a random subset of caregivers. The resulting descriptive data provided insight into how children on the autism spectrum experience and communicate pain to the world around them.

**Study Design**

This two-phase study was approved by the University of Florida Health Science Center, Institutional Review Board (IRB). During Phase I, a survey was developed by the author and mailed to four hundred families of children with a diagnosed autism spectrum disorder. During Phase II, ten caregivers participated in a semi-structured phone interview in order to discuss their child’s pain expression in more detail.

**Participants: Recruitment, Consent, and Anonymity**

Participants included caregivers of children with a diagnosis of autism, PDD-NOS, or Asperger’s disorder (AD). All caregivers were identified as primary caregivers, living in the home with their child with ASD, and all were able to write and speak in English. Surveys were distributed, by mail, to 400 caregivers whose children utilized services and resources provided by the Center for Autism and Related Disabilities (CARD) at the University of Florida. Response was voluntary. A detailed cover letter provided information regarding the study aims and caregiver rights as participants (see Appendix A). This study was conducted with IRB
permission to waive documentation of informed consent, under the assumption that by voluntarily responding, it could be safely assumed that participants implied their consent.

Interested caregivers anonymously responded by completing the mailed survey and returning it (also by mail) in a pre-addressed pre-stamped envelope, which had been included with the survey. Caregivers were given the option to provide their first name and telephone number, if they were interested in participating in the semi-structured interview via a follow up phone call. A subset of caregivers were randomly selected, and then invited to participate in Phase II. Telephone interviews were then conducted. Caregiver responses and thoughts were hand-written as the interview was conducted, and then typed by the primary investigator. All surveys and response sheets were de-identified prior to analysis.

**Measures/Instruments**

Two instruments were developed for this exploratory descriptive study, the “Caregiver Survey on Pain Expression in Children with ASD” and the “Semi-Structured Interview Guide to Discuss Pain in ASD” (see Table 3-1).

**Caregiver Survey on pain expression in children with ASD**

The Caregiver Survey was developed in order to elicit perceptions caregivers have regarding their child’s pain expression. In addition to basic demographic information, participants were asked three “yes” or “no” questions, and were prompted to provide additional descriptive information regarding their child.

**Semi-structured interview guide to discuss pain in ASD**

Almost 80% of survey respondents provided a first name and phone number in order to receive a follow-up call. A random set of ten caregivers were called to conduct the semi-structured interview. The interview involved asking ten open-ended questions with each
participant. Caregivers were allowed to elaborate on those issues that they felt were most important and concerning to them.

**Data Synthesis and Analysis**

Caregiver survey responses were compiled. Answers to “yes” or “no” questions were quantified, and descriptive information was examined to identify common themes and issues. All pain expressive indicators (or behaviors) described by caregivers were noted, and used to generate a growing list of potential pain indicators in this population. This list of items was utilized in Study 2 in order to determine whether it is possible to develop a pain assessment tool for use in this population. Caregiver challenges regarding pain assessment in their children were noted. Details from the semi-structured interviews were used to better qualify the beliefs and perspectives held by caregivers regarding pain expression in their children. Several case studies were drafted to inform others about pain expression in children with ASD.

**Study 2: Expert Appraisal of Potential Pain Expressive Indicators in ASD**

The second study conducted in this research was titled “Expert-Rated Relevancy of Potential Pain Expressive Indicators in ASD: Establishing Content Validity.” From this point on, this second study will be referred to as “Study 2.”

**Purpose**

The purpose of this study was to identify a core set of indicators relevant to pain assessment in ASD. This was accomplished by a) generating a comprehensive list of potential pain expressive indicators in children with ASD, b) having this list reviewed by autism experts, and c) establishing the content validity of each item in order to reduce the list to a number of items achieving an acceptable scale-level content validity.
Participants: Expert Reviewers

Fifteen experts were invited to participate in this study. Experts were identified first by the investigator, and then discussed with a seasoned autism expert—whose role was to further examine and validate the background and experience of the identified experts. Experts needed to be currently working with children diagnosed with ASD. They needed to have a history of ongoing clinical and/or research projects in the fields of autism and/or pain assessment in vulnerable populations of children. Of the fifteen experts invited to participate, it was estimated that between six and nine would be able to review the list of potential pain indicators, which is the number necessary to establish content validity for individual items and the resulting scale.

Measures/Instruments: Item List for Expert Review

It is universally recommended that a large item pool be generated when developing instruments, in order to allow an investigator to include all possible items “tapping into” a construct, and thus devise a psychometrically strong instrument. The definition of “large” varies from one and a half times as large as the anticipated final scale (Allen & Yen, 1979) to three or four times as large as the final version (DeVellis, 2003). Current pain assessment tools for use in specialized populations, such as the Non-Communicating Children’s Pain Checklist, are about 30 items long. Preliminary reports indicate that this tool with 30 items is easy to use by caregivers, takes little time, and accurately discriminates pain in populations in which it has been validated (Breau, Finley, McGrath, & Camfield, 2002; Breau, McGrath, Camfield, & Finley, 2002). Given that, theoretically, several of the items in this already developed tool might be appropriate for children with ASD, while other items may need to be removed, or replaced, it was anticipated that the final version of the tool in development might be between 20 and 30 items. Review of the pain and autism literature, examination of other observational pain assessment tools,
caregiver surveys and interviews from Study 1, and preliminary discussions with autism experts—none of whom participated in Study 2—generated 77 initial items.

An item list was developed that consisted of these 77 behaviors that might indicate pain in children with ASD (see Appendix B). This total number of initial items was appropriate, given the recommendations that the item pool be somewhere between one and a half (45) and four times (120) larger than the anticipated final instrument. These behaviors were put into 6 categories: “Vocal/Verbal Indicators,” Physiological Indicators,” “Activity/Movement,” “Behavioral Indicators,” “Daily Activities,” and “Facial Indicators.”

**Procedures and Data Analysis**

An initial iteration of the content validity process was conducted with experts in the fields of autism and pain to closely examine the current item pool. Items were revised, removed, or retained based on expert input regarding the reported relevance to pain assessment in children with ASD.

**Establishing content validity**

Measurement experts emphasize the importance of establishing content validity. Content validity represents “the degree to which elements of an assessment instrument are relevant to and representative of the targeted construct for a particular assessment purpose” (Haynes, Richard, & Kubany, 1995, p. 238). Throughout the literature, researchers commonly report that “content validity was established,” however the process by which content validity was established is not always articulated. It is important to point out that establishing content validity is a process. As articulated by Polit, Beck and Owen (2007), it may be necessary to undergo several iterations of item review, especially if the first review reveals that a number of item revisions, clarifications, or additions are warranted. This research aimed to complete the first iteration of item review, and implications for a second round with experts are discussed in Chapter 5.
Before beginning this study, various methods for establishing content validity were examined. The process for establishing content validity has been described in several nursing texts, and is further outlined in a number of articles (Polit & Beck, 2006; Polit, Beck, & Owen, 2007). One of the commonly reported quantitative measures of content validity is the content validity index (CVI). Lynn (1986; as cited in Polit & Beck, 2006) recommends that between three and ten experts participate in the content validity process. Each expert rates every item on a 4-point ordinal scale labeled: 1 (not relevant), 2 (somewhat relevant), 3 (quite relevant), and 4 (highly relevant). Following review by all experts, an item-level CVI (I-CVI) is calculated for each item. For this study, a cover letter describing the research accompanied the item list (see Appendix C) and instructed the reviewers to rate each of the 77 items as either “not relevant,” “somewhat relevant,” “quite relevant,” or “highly relevant” to pain assessment in children with ASD.

**Item-level content validity indexes (I-CVI)**

I-CVI is calculated by figuring the “number of experts giving a rating of either three or four…divided by the total number of experts” (Polit & Beck, 2006). To accommodate for chance agreement, Lynn (1986; as cited in Polit & Beck, 2006) advocates that if there are five or fewer judges, the I-CVI should be 1.00 in order to retain an item as relevant. For six or more judges, she recommends an I-CVI of .78 or above. Others have calculated the I-CVI with five judges and accepted any item with an I-CVI above .80 (Decker & Perry, 2003). Polit, Beck, & Owen (2007) state that because establishing content validity may require several expert reviews, in the initial iteration it is wise to retain items with an I-CVI of .78 or above, however it may be acceptable to consider items with an I-CVI somewhat lower than .78 as candidates for revision—rather than discard them too hurriedly. Lynn’s (1986) methods are most commonly reported throughout the nursing literature, and have been adopted by many researchers embarking on
instrument development. Her methods are sometimes critiqued because they do not statistically take into account the possibility for “chance agreement” (Wynd, Schmidt, & Schaefer, 2003). It is pointed out that other indicators are available to calculate interrater agreement (kappa, for example), and do account for chance agreement (Polit, Beck, & Owen, 2007; Wynd, Schmidt, & Schaefer, 2003). Supporters of the CVI counter argue that kappa accounts for chance agreement and chance disagreement. Kappa also does not provide information at the item-level—which is often necessary when examining each item’s relevance (or irrelevance) to a particular assessment tool.

I-CVIs provide information that decision-makers need in order to construct an assessment scale. Polit, Beck, and Owen (2007) argue that the CVI is appropriate for item-level analysis, and to address the debate regarding “chance agreement,” they calculated a modified kappa statistic (taking into consideration both chance agreement and the proportion of expert agreement on relevance) to allow an investigator to compare each calculated I-CVI with what would translate into an “excellent,” “good,” or “fair” modified kappa coefficient. To articulate “excellent,” “good,” or “fair,” interrater agreement, they utilize the kappa coefficient criteria set by Cicchetti and Sparrow (1981). In a sense, their calculations indicate that items with high I-CVIs often have adequate modified kappa coefficients, therefore indicating that the I-CVI is useful even with the described limitations regarding chance agreement (and disagreement).

This current research utilized the table developed by Polit et al. (2007) to determine whether all calculated I-CVIs were “excellent,” “good,” or “fair” in relation to their modified kappa. For this study, it was established a priori that six experts would participate in the initial expert review, and a cut-off of .78 would be utilized to determine whether items should be
retained. Items with an I-CVI just below .78 would be considered for revision, and those with the lowest I-CVIs would be removed from the list.

**Scale-level content validity index**

Despite use of the I-CVI to inform scale developers whether to discard or retain items, many published articles describing various scales’ development only report the scale-level CVI (S-CVI). Because there are several ways to compute the S-CVI, it is important for authors to provide information on how the S-CVI was computed (Polit & Beck, 2006). Polit and Beck (2006) recommend calculating the S-CVI by reporting the *average* of all the I-CVIs in a final scale. For example, a scale with five items, whose I-CVIs are 1.0, 1.0, 1.0, .83, and .83, would have a calculated S-CVI of \( \frac{1+1+1+.83+.83}{5} = .93 \). This calculation is referred to as the “S-CVI/Ave” by Polit & Beck (2006). There is debate regarding whether the acceptable S-CVI should be set at .80 or .90 (Polit & Beck, 2006).

This initial item review with experts revealed that it was possible to remove some items, retain some items, and that there were some items that needed to be revised. As previously discussed, Polit, Beck, and Owen (2007) point out that it may be necessary to undergo multiple iterations in an effort to establish content validity. Because the initial list of items needed revision, an S-CVI was not yet calculated. If it is determined, following another review by experts, that item revision is no longer necessary; the S-CVI/Ave will be calculated according to specifications outlined by Polit and Beck (2006). An a priori determination will be made to accept .80 as an acceptable scale-level content validity.

Content validity procedures guarantee that the theoretical conceptualization of the investigator is supported by those considered to be experts in the field. This process ensures that items remaining in the tool following these content validity procedures are a) supported
theoretically by the literature, b) supported by qualitative and/or quantitative study findings and/or clinical experience, and c) have systematically undergone expert review.

**Expert comments and suggestions**

Expert suggestions, questions, and comments were solicited following completion of the items list review. Experts were given the opportunity to comment on specific items, or perhaps to discuss the overall process of this instrument’s development. Many researchers embarking on instrument development underutilize the expert advice that is available (Grant & Davis, 1997). Careful thought was given in order to select experts that would be interested in the current research, so that their advice and expertise would be readily available throughout the process. Expert comments and suggestions resulting from the item review have been thoughtfully considered and are reported in Chapter 5. The implications of their thoughts on this instrument’s further development will also be discussed.

**Summary**

The methods presented here describe an initial attempt to think about the long-term nature of understanding how to assess pain in children with ASD. It is the intent of this discussion to ensure readers that the writer is adequately familiar with the various issues related to instrument development—designing studies that will ensure the development of psychometrically sound instruments, understanding reliability and validity, and how to establish such measures, etc. Thoughtful preparation for every step in the process of instrument development, including the two studies conducted for this research, ensures the development of a psychometrically sound tool for assessing pain in children with autism spectrum disorder.
<table>
<thead>
<tr>
<th>Table 3-1. Study 1: Caregiver survey and semi-structured interview questions</th>
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<tr>
<td><strong>Caregiver survey</strong></td>
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<tr>
<td>Compared to other children your child’s age, do you feel your child responds differently to painful events? Please describe.</td>
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<tr>
<td>Do you find it difficult to tell when your child might be in pain? Please describe.</td>
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<tr>
<td>Do you feel healthcare providers are able to assess and treat your child’s pain well? Please describe.</td>
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CHAPTER 4
RESULTS

The findings from two studies are presented in this chapter. First presented are results from Study 1, including demographic information, and quantitative data for each question. Then, descriptive information provided by caregivers—from surveys and semi-structured interviews—is presented. Next, results of Study 2 are described, beginning with item-level analysis. Items achieving acceptable content validity are presented. Those items not achieving an I-CVI of .78 are also discussed. Thoughts and comments provided by expert reviewers are reported, and anticipated item list revisions are introduced, but are more thoroughly discussed in Chapter 5.

Study 1: Caregiver Survey Results

Eighty-eight surveys were completed by caregivers of children with ASD and returned by mail. All surveys were included in data analysis. This is equivalent to a return rate of 22%. Given this low response rate, it is important to consider how a high nonresponse rate might influence findings and subsequent conclusions. This is discussed further in the Limitations section of Chapter 5.

Demographic Information

Caregivers reported that their children had a diagnosis of either autism (n = 32), PDD-NOS (n = 19), Asperger’s disorder (n = 12) or “likely autism” (n = 25). For the purposes of this study, a category “likely autistic” was used because some children exhibited enough autistic behaviors to receive services from the Center for Autism and Related Disabilities, but the child’s official ASD diagnosis may not have been conclusively established at the time the survey was completed. A majority of the children described in Study 1 were autistic or likely autistic (n = 57). The population of children described by caregivers consisted of 62 boys, 20 girls, and 6
children whose gender was not reported. These numbers reflect reported trends that ASD occurs three times more often in boys than girls. For this sample, the male female ratio was 3.3:1 (62 boys, 19 girls).

Quantitative Results from Caregiver Surveys

Table 4-1 summarizes the quantitative responses to the three survey questions. Eighty-six percent (n = 76) of caregivers felt their child responded differently to pain. Seventy percent (n = 62) stated they find it difficult to determine when their child might be in pain, and fifty-six caregivers (64%) responded that healthcare providers either did not—or only sometimes—adequately assessed and treated their child’s pain.

Descriptive Information Provided by Caregivers

Question 1: Compared to other children your child’s age, do you feel your child responds differently to painful events? Please describe. Written caregiver responses provided descriptive information regarding pain expression in children with ASD. The responses to Question 1 were examined, and themes were identified by the author and an autism expert. Themes common to those caregivers who do feel their child responds differently to pain are reported. These themes highlight the most commonly reported differences articulated by caregivers, and were identified by looking at how frequently each issues was mentioned. The themes, along with a description/definition of each, are summarized in Table 4-2, and include “exhibits altered sensitivity to pain,” “displays unusual/unique pain expression,” “demonstrates pain seeking behavior,” “pain expression varies by type,” and “pain expression varies with development.”

The theme, “exhibits altered sensitivity to pain,” refers to a variety of caregiver reports describing either hyper-sensitivity or hypo-sensitivity to pain. Several caregiver quotations illuminating this commonly reported phenomenon are presented in Table 4-3. The theme,
“displays unusual/unique pain expression,” refers to those responses where caregivers correlate specific behaviors and/or indicators to pain in their children. They openly acknowledged that these reported pain indicators are not typically associated with pain in children. Finally, examples of caregiver quotes from which the themes “demonstrates pain seeking behavior,” “pain expression varies by type,” and “pain expression changes with development” were derived are also provided in Table 4-3.

**Question 2: Do you find it difficult to tell when your child might be in pain? Please Describe.** Caregivers were asked whether they had difficulty determining when their child might be in pain. Seventy percent (n = 62) stated they find it difficult to determine when their child might be in pain. Written descriptions (n = 39, 44%) of the difficulties associated with pain assessment primarily reflected challenges related to working with nonverbal children (and thus children who do not verbally complain). Other caregivers (n = 27, 31%) reported that because their children do not cry, or because they appear stoic, it is difficult to determine if they are in pain.

The thirty percent (n = 26) of caregivers reporting that it is not difficult to determine when their children are in pain stated that their children were “verbal” (n = 10), or they could identify specific pain expressive behaviors unique to their child. These reported behaviors are listed in Table 4-4, and were referenced when generating the comprehensive list of potential pain expressive behaviors for review in Study 2. Other caregivers (n = 4) stated they can tell when something is wrong, but get frustrated when their child cannot help them determine “where” or “how serious” the pain is.

**Question 3: Do you feel health care providers are able to assess and treat your child’s pain well? Please describe.** Fifty-six caregivers (64%) reported that healthcare
providers either did not, or only sometimes, adequately assessed and treated their child’s pain. Written responses highlighted that many parents believe pain is an indicator that something is wrong in the body. Because they have a difficult time determining when their child is in pain, they are forced to depend on other signals (i.e., fever, gait disturbances) to alert them that something might be physically wrong. The prevalent caregiver concerns involved their ability, as caregivers, to advocate for their children. They reported not being taken seriously at healthcare facilities when they knew “something was wrong,” but their child was not exhibiting any typical pain expressive behaviors. A number of parents reported that they have worked hard to find primary care physicians who understand ASD, and who will advocate for them and their children within the healthcare system.

**Study 2: Expert-Rated Relevance of Potential Pain Indicators**

Information gleaned from Study 1 characterized caregiver perceptions regarding the pain experience in children with ASD, and highlighted challenges associated with attempting to assess pain in children with social and communicative impairments. Caregiver-identified pain indicators were added to a growing list of expressive/observable behaviors that might indicate pain in children with ASD. In Study 2, this list was reviewed by autism and pain experts in order to a) gain additional insight into pain expression in children with ASD, and b) identify behaviors considered highly relevant to pain assessment in ASD.

**Examination of Item-Level Content Validity Indexes**

An item-level content validity index (I-CVI) was computed for each of the 77 items reviewed by the expert panel. I-CVIs ranged from 0 (where not one expert believed the item was either “quite” or “highly” relevant, to 1.0 (where ALL experts believed the item was “quite” or “highly” relevant to pain assessment in ASD. The calculated I-CVI for each item is displayed in Table 4-5. In summary: 4 items had an I-CVI of 0, 13 items had an I-CVI of .17, 13 items had
an I-CVI of .33, 14 items had an I-CVI of .5, 19 items had an I-CVI of .67, 13 items had an I-CVI of .83, and 1 item had an I-CVI of 1.0.

For this study, it was established a priori that six experts would participate in the expert review, and an I-CVI cut-off of .78 would be utilized to determine whether items should be retained, revised, or removed from the list. Using these guidelines, 14 items achieved an I-CVI greater than .78. These items are referred to as the Primary Pain Indicators and achieved a high enough I-CVI to remain in the developing pain assessment tool. These items include: “rocking unusually,” “lashing/striking out,” “being less active,” “banging his/her head,” “injuring oneself,” “crankiness (being more cranky than usual),” “lying around/acting lethargic,” “increased heart rate,” “furrowing brow,” “screaming/yelling/throwing tantrums,” “guarding (protecting) or favoring a particular part of the body when moving,” “inconsolability (being unable to console),” “tensing face,” and “grimacing.” Five of these items were originally categorized as “Activity/Movement” pain indicators. Three items were from the “Behavioral” category, three from the “Facial” category, one from “Vocal/Verbal,” one from “Daily Activities,” and one from “Physiological.” This highlights that experts considered a range of indictors to be relevant to pain assessment, from each of the aforementioned categories.

As recommended by Polit, Beck, and Owen (2007), the group of indicators that achieved an I-CVI of .67 (just below the pre-established I-CVI = .78) were not completely removed at this point in the instrument’s development, and will be considered for revision. They are referred to as the Secondary Pain Indicators (n = 19), and the implications of revising, removing, or retaining each of these items are discussed in Chapter 5. These items include: “acting ‘off’ according to caregiver, not acting like him/her self,” “crying,” “moaning, whining, whimpering (softly),” “stating/repeating a specific pain-related word,” “seeking comfort or physical touch,”
“touching rubbing, picking and/or tapping a specific part of the body,” “changing color (flushing/redness),” “producing tears (without overt crying or vocalizations),” “increased rate of breathing,” “sleeping less,” “grinding teeth, clenching jaw down,” “frowning, turning down mouth,” “pouting, quivering, puckering lips,” “widening eyes,” “becoming more aggressive (hitting, pinching, biting others,” “acting angry,” and “holding his/her head for prolonged periods of time.”

**Calculation of a Scale-Level Content Validity Index**

Because this expert review resulted in the need to revise items—and perhaps re-categorize the items rated most relevant to pain assessment in ASD—the final form of this instrument was not yet realized. It was also therefore, not appropriate to calculate the S-CVI at this point in the instrument’s development. This decision is addressed again in the Chapter 5. Once a set of relevant items are finalized—and prior to additional administration of the instrument to the sample population—the S-CVI will be calculated.

**Expert Comments and Suggestions**

Expert comments are briefly mentioned here, and the implications of these thoughts and suggestions are discussed in Chapter 5. Comments generally included a) suggestions regarding how to re-word particular items, b) requests that better/additional operational definitions for particular items be provided, c) suggestions on how to more clearly categorize the relevant pain indicators, d) information on how they (the particular expert) rated whether items were considered relevant or not, and e) suggestions for the next steps in this instrument’s development.

**Summary**

These results highlight the perceptions held by caregivers regarding the pain experience in children with ASD. Their insights regarding the indicators they use to determine when their
children are in pain demonstrate the unique nature of pain expression in this population. These indicators were added to the list of potential pain indicators that had been already derived from the pain and autism literature. Finally, expert review of the entire list identified key pain expressive behaviors that will be used in the further development of an instrument to assess pain in children with ASD.
### Table 4-1. Study 1: Quantitative results from caregiver surveys

<table>
<thead>
<tr>
<th>Question</th>
<th>Yes</th>
<th>No</th>
<th>Sometimes</th>
<th>Don’t know</th>
</tr>
</thead>
<tbody>
<tr>
<td>Compared to other children your child’s age, do you feel your child responds differently to painful events?</td>
<td>76 (86%)</td>
<td>12 (14%)</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Do you find it difficult to determine when your child is in pain?</td>
<td>62 (70%)</td>
<td>26 (30%)</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Do you feel healthcare providers are able to assess and treat your child’s pain well?</td>
<td>19 (22%)</td>
<td>40 (45%)</td>
<td>16 (18%)</td>
<td>13 (15%)</td>
</tr>
</tbody>
</table>

Note: n=88
<table>
<thead>
<tr>
<th>Identified themes</th>
<th>Detailed description/definition</th>
</tr>
</thead>
<tbody>
<tr>
<td>Exhibits altered sensitivity to pain:</td>
<td></td>
</tr>
<tr>
<td>Hyper-sensitive to pain</td>
<td>Child exhibits <em>exaggerated</em> response to minimal or seemingly non-painful events</td>
</tr>
<tr>
<td>Hypo-sensitive to pain/has a high tolerance for pain</td>
<td>Child exhibits a <em>minimal</em> or <em>lack of</em> response to seemingly painful event</td>
</tr>
<tr>
<td>Hyper-hypo-sensitive phenomenon</td>
<td>Child <em>oscillates</em> between over-reacting and under-reacting to seemingly painful events</td>
</tr>
<tr>
<td>Displays unusual/unique pain expression</td>
<td>Child demonstrates <em>delayed</em> response to pain, or exhibits unusual/atypical pain expressive behaviors as identified by caregiver (i.e., laughing)</td>
</tr>
<tr>
<td>Demonstrates pain seeking behavior</td>
<td>Child engages in self-injurious behavior or <em>seeks out</em> painful Stimuli</td>
</tr>
<tr>
<td>Pain expression varies by <em>type</em></td>
<td>Child reacts differently to various <em>types</em> (acute vs. chronic) or <em>degrees</em> of pain</td>
</tr>
<tr>
<td>Pain expression changes with development</td>
<td>As children grow and develop, pain expression and the ability to communicate pain changes</td>
</tr>
<tr>
<td>Caregiver quote</td>
<td>Identified theme</td>
</tr>
<tr>
<td>-------------------------------------------------------------------------------</td>
<td>----------------------------------------------------------------------------------</td>
</tr>
<tr>
<td>“Benjamin* is very demonstrative…he seems to feel things more intensely.”</td>
<td>Exhibits altered sensitivity to pain: Hyper-sensitivity</td>
</tr>
<tr>
<td>“He has extreme dramatic reactions to unexpected pain. Loud screaming and</td>
<td></td>
</tr>
<tr>
<td>tantrums. It takes some time to calm him down.”</td>
<td></td>
</tr>
<tr>
<td>“He makes a big thing about something small, like cutting his finger a little bit.”</td>
<td></td>
</tr>
<tr>
<td>“It takes a while for her to feel pain. Sometimes has no pain…she can get hit or bump into things and not realize she is hurt or bleeding.”</td>
<td>Exhibits altered sensitivity to pain: Hypo-sensitivity or high tolerance for pain</td>
</tr>
<tr>
<td>“My child only responds to severe pain and then he only rubs the place that has been injured and briefly fusses. Most times he doesn’t even acknowledge his injuries.”</td>
<td></td>
</tr>
<tr>
<td>“A light touch will sometimes cause a painful reaction. Whereas, a deeper touch will not. Minor pains will cause crying. More moderate to severe pains will sometimes cause laughter!”</td>
<td>Exhibits altered sensitivity to pain: (Hyper-hypo-sensitive phenomenon)</td>
</tr>
<tr>
<td>“My daughter will either overreact or show no signs that it was painful…for example I noticed she had half of one toenail gone…but she never reacted to it at all…Other times she will go on and on about something as small as a little scratch.”</td>
<td></td>
</tr>
<tr>
<td>“Generally, my child does not complain of minor pains. He will punch himself in the stomach and that indicates he is a little constipated….”</td>
<td>Displays unusual/unique pain expression</td>
</tr>
<tr>
<td>“He doesn’t talk. If he is in pain he might stomp his foot or clap his hands loud.”</td>
<td></td>
</tr>
<tr>
<td>“Just sort of ‘off’ and less active and cheerful.”</td>
<td></td>
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<tr>
<td>“She will say odd things like ‘it tickles’. ”</td>
<td></td>
</tr>
<tr>
<td>“Often giggles instead of saying ouch.”</td>
<td></td>
</tr>
<tr>
<td>Caregiver quote</td>
<td>Identified theme</td>
</tr>
<tr>
<td>-------------------------------------------------------------------------------</td>
<td>------------------------------------------------------</td>
</tr>
<tr>
<td>“Jon* enjoys putting hands on hot things, such as light bulbs and oven burners.”</td>
<td>Demonstrates pain seeking behavior</td>
</tr>
<tr>
<td>“She will moan and bite her hand.”</td>
<td></td>
</tr>
<tr>
<td>“He under and over reacts...and he often seeks out ways to hurt himself—throwing himself on the floor, biting his own arms.”</td>
<td></td>
</tr>
<tr>
<td>“Blunt pain isn’t as noticeable to him. However a splinter or scrape to the skin is very excessive in his expression.”</td>
<td>Pain expression varies by type</td>
</tr>
<tr>
<td>“Yes to trauma types, i.e., needle pricks...no to gas headaches, small cuts, blisters on his feet”</td>
<td></td>
</tr>
<tr>
<td>“When he was younger, he did not respond to pain. For example, one day he had new shoes on all day...and I didn’t know they had the paper stuffed in them! Or he’d bang his head and bite his hands. Now he’s eight and can talk well, but now it seems he over-reacts to pain. The tiniest little thing gets him very upset.”</td>
<td>Pain expression changes with development</td>
</tr>
<tr>
<td>“He reacts and seems to feel pain more now than when he was younger.”</td>
<td></td>
</tr>
<tr>
<td>“He has improved greatly in his ability to recognize and describe pain...only in the past two years has he been able to tell me he has a headache.”</td>
<td></td>
</tr>
</tbody>
</table>

Note: * indicates names have been changed.
<table>
<thead>
<tr>
<th>Behavioral indicators</th>
<th>Activity/Movement</th>
<th>Vocal/Verbal indicators</th>
<th>Changes in routine</th>
</tr>
</thead>
<tbody>
<tr>
<td>Lashes out/becomes aggressive</td>
<td>His/her body language Changes</td>
<td>Cries</td>
<td>Lays around more</td>
</tr>
<tr>
<td>“Uses actions”</td>
<td>Bangs his/her head</td>
<td>Screams/throws tantrums</td>
<td>Sleeps more</td>
</tr>
<tr>
<td>“Stomps his foot”</td>
<td>“Touches whatever hurts”</td>
<td>Gets quiet</td>
<td>“Something is just off”</td>
</tr>
<tr>
<td>“Claps his hands loud”</td>
<td>Holds his/her head</td>
<td>“Utters/states a specific word”</td>
<td>Is less cheerful</td>
</tr>
<tr>
<td>“Bites his hand”</td>
<td>“Hunches over when walking”</td>
<td>Laughs</td>
<td>Is less active</td>
</tr>
<tr>
<td>“Rocks violently”</td>
<td>“Hurts himself”</td>
<td>Moans</td>
<td>“Is listless”</td>
</tr>
<tr>
<td>Hides</td>
<td>“Gets still”</td>
<td>“He tells me”</td>
<td></td>
</tr>
</tbody>
</table>

Note: Indicators in quotation were taken directly from caregiver written responses. Indicators not in quotation were derived by the investigator to summarize concisely what several caregivers reported.
<table>
<thead>
<tr>
<th>I-CVI = 0</th>
<th>I-CVI = .17</th>
<th>I-CVI = .33</th>
<th>I-CVI = .5</th>
<th>I-CVI = .67</th>
<th>I-CVI = .83</th>
<th>I-CVI = 1.0</th>
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</thead>
<tbody>
<tr>
<td>Humming/ singing</td>
<td>Interacting more than normal with others</td>
<td>Decreasing number and/or frequency of repetitive behaviors</td>
<td>Sleeping more</td>
<td>Sleeping less</td>
<td>Lashing/striking out</td>
<td>Rocking unusually</td>
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<tr>
<td>Laughing</td>
<td>Rolling eyes</td>
<td>Sitting still</td>
<td>Feeling cool or cold to touch</td>
<td>Irritability (being more irritable than usual)</td>
<td>Being less active</td>
<td>—</td>
</tr>
<tr>
<td>Eating more than usual</td>
<td>Raising eyebrows</td>
<td>Stomping foot</td>
<td>Lip biting</td>
<td>Seeking comfort/physical touch</td>
<td>Banging his/her head</td>
<td>—</td>
</tr>
<tr>
<td>Stating/repeating a specific non pain word or phrase (i.e., bat, toe)</td>
<td>Decreased rate of breathing</td>
<td>Perseverating more on a particular item</td>
<td>Fever</td>
<td>Becoming more aggressive (i.e., hitting, pinching, biting others)</td>
<td>Injuring oneself</td>
<td>—</td>
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<tr>
<td>—</td>
<td>Seeming less sensitive to touch</td>
<td>Making up stories or reenacting a painful circumstance</td>
<td>Scratching a specific part of the body</td>
<td>Acting angry</td>
<td>Crankiness (being more cranky than usual)</td>
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<tr>
<td>—</td>
<td>Becoming floppy, or “goes limp”</td>
<td>Lip curling</td>
<td>Interacting less than normal with others</td>
<td>Widening eyes</td>
<td>Laying around, acting lethargic</td>
<td>—</td>
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<tr>
<td>—</td>
<td>Eating less, not interested in food</td>
<td>Fidgeting, becoming more active, jumping around (motor restlessness)</td>
<td>Isolating oneself more, hiding</td>
<td>Frowning, turning down mouth</td>
<td>Increased heart rate</td>
<td>—</td>
</tr>
<tr>
<td>I-CVI = 0</td>
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<tr>
<td>—</td>
<td>Holding breath</td>
<td>Cooperating less than normal</td>
<td>Loosing interest in things that are usually extremely important or interesting to child</td>
<td>Pouting, quivering, puckering lips</td>
<td>Furrowing brow</td>
<td>—</td>
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<tr>
<td>—</td>
<td>Chewing, moving tongue around and/or out, smacking lips</td>
<td>Facial restlessness, tics</td>
<td>Increasing number and/or frequency of repetitive behaviors</td>
<td>Producing tears (without overt crying or vocalizations)</td>
<td>Screaming/yelling/throwing tantrums</td>
<td>—</td>
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<tr>
<td>—</td>
<td>Babbling (saying something incomprehensible)</td>
<td>Vocalizing less than normal, getting quiet</td>
<td>Flaring nostrils</td>
<td>Increased rate of breathing</td>
<td>Guarding (protecting) or favoring a particular part of the body when moving</td>
<td>—</td>
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<td>—</td>
<td>Clapping hands loudly</td>
<td>Seeming hypersensitive (overly sensitive) to touch</td>
<td>Muscle tensing, rigidity</td>
<td>Acting “off” according to caregiver; not acting like him/herself</td>
<td>Inconsolability (being unable to console)</td>
<td>—</td>
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<td>—</td>
<td>Continuous eye blinking</td>
<td>Shivering, Shaking</td>
<td>Unhappiness</td>
<td>Grinding teeth, clenching jaw down</td>
<td>Tensing face</td>
<td>—</td>
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<tr>
<td>—</td>
<td>Moving to, or staying in, a specific location or “safe” space</td>
<td>Changing his/her usual body language</td>
<td>Sweating, perspiring</td>
<td>Squinching, squinting eyes</td>
<td>Grimacing</td>
<td>—</td>
</tr>
<tr>
<td></td>
<td>I-CVI = 0</td>
<td>I-CVI = .17</td>
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<td></td>
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<td></td>
<td></td>
<td>Changing color (paleness)</td>
<td>Crying (with or without tears)</td>
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<td></td>
<td></td>
<td></td>
<td>Moaning, whining, whimpering (softly)</td>
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<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Changing color (flushing, redness)</td>
<td></td>
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<td></td>
<td>Touching, rubbing, picking, and/or tapping a specific part of the body</td>
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<td></td>
<td>Holding his/her head with both hands for prolonged periods of time</td>
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<td></td>
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<td></td>
<td></td>
<td></td>
<td>Stating or repeating a specific pain-related word (i.e., hurt, ouch)</td>
<td></td>
</tr>
</tbody>
</table>

Note: * I-CVI was calculated by looking at the proportion of expert reviewers rating an item’s relevancy as either “quite” or “high.” For example, if five of six experts rated an item as either “quite” or “highly” relevant to pain assessment in ASD, the calculated I-CVI would be 5/6 = .83. All items in the “I-CVI = .83” column had a calculated I-CVI of .83.
CHAPTER 5
DISCUSSION

This research explored pain expression in children with autism spectrum disorder (ASD). These two studies add to what is known about how vulnerable pediatric populations—specifically, those unable to accurately self-report pain—express their discomfort to those around them. This research focused on children with autism spectrum disorder, whose pain experience has hardly been investigated. The literature review conducted for this research revealed one empirical study examining pain in this population (Nader et al., 2004). Nader et al. (2004) demonstrated that children with ASD reflexively (involuntarily) respond to painful stimuli, indicating that perhaps the perceptions of pain insensitivity in this population are inaccurately founded. This dissertation research extended those findings, to better describe what caregivers and autism experts—presumably, individuals who know this population best—believe about how these individuals experience and express pain. Finally, this research facilitated the initial development of a pain assessment tool for use in this population, by identifying specific pain indicators relevant to children with ASD.

Pain Expression in ASD Compared to Typically Developing Children

Nader et al. (2004) reported that a) caregivers of children with ASD might be confused about pain in their children, and that b) parents perceive insensitivity to pain in their children when compared to nonimpaired children. Findings in this research are consistent with these reports. Caregivers stated that they do believe their children respond differently to pain when compared to other developing children. When asked to qualify these differences, however, they did not always refer to their children as having “a high tolerance for pain,” or “insensitivity to pain.” In this way, these findings are new and unique, adding to what is known about how caregivers perceive the pain experience in their children.
Several caregivers did refer to their children as having a minimal reaction to everyday painful occurrences; however a majority of caregivers described a phenomenon whereby their child would attend to various types of pain differently. They described how a deep cut on the back of the child’s head would cause minimal discomfort, but a tiny scratch on the hand would result in him or her being hysterical. This phenomenon seems to be somewhat unique to this population. Bursch, Ingman, Vitti, Hyman, & Zeltzer (2004) posit that children with ASD may have intact acuity to sensory input; however their processing of incoming information might be distorted. Thus “rain might sound like gunfire, clothing might feel like sandpaper, or fingers shampooing a scalp might feel like sharp metal” (p. 290). These sensory abnormalities have potential to distort the processing of pain stimuli as well, speaking to the fact that they feel the stimulus, but they may not attend to it as other children do. Thus, the reports by caregivers that their children seem hypersensitive to some pain, and then hyposensitive to other pain, might be explained by this overall abnormal sensory processing.

This “unusual responsiveness to the environment can be explained in part by stimulus overselectivity, the tendency…to respond to only a very limited amount of relevant sensory information” (Bursch et al., 2004, p. 291). One caregiver described a situation where her child would grab a hot curling iron and not respond as if he were in pain, but yet he would scream and yell in “pain” from the tag in his shirt. Sensory abnormalities are not seen in every child on the autism spectrum, but it may be that those children with sensory abnormalities process—focus on different aspects of—pain sensations differently. Additional investigation into subgroups of children with ASD, for example those with sensory abnormalities, is warranted.

Caregiver comments on how their children, when they did report pain, would perseverate on the pain (or source of pain) also highlight differences in the pain experience in this population.
compared to typically developing children. The perseverative tendencies unique to children with ASD permeate all types of experiences, including their pain experience. Bursch et al. (2004) stated that “once focused on pain, difficulties in shifting attentional focus can serve to increase pain and associated distress” (p. 294). Interrupting the perseveration then, might lead to pain relief. Zeltzer and Schlank (2005) describe a situation in their pain clinic where a child with ASD presented to them yelling repeatedly, “Ow! Ow! Ow!” He had been shouting for the past two months, since sustaining a leg injury. Zeltzer’s immediate suggestion that he squeeze a ball instead of shouting “Ow!” resulted in a transfer of his expression, and he actually reported feeling better because he was no longer embarrassed about shouting “Ow!” all the time. While this particular perseverative response was clearly associated with pain (i.e., it is easy to infer the child was in some sort of pain because the word “Ow!” is typically associated with pain), one has to consider that other children may also engage in repetitive behaviors when in pain—however they may select (especially if they are unable to verbally communicate) other behaviors not typically associated with pain (i.e., “rocking unusually”). These articulated differences indicate that caregivers do not always believe their children are incapable of feeling pain.

**Determining When Children with ASD are in Pain**

Caregivers reported that they find it difficult to determine when their children are in pain. Of those caregivers who can determine when their children are in pain, many are frustrated because they can never figure out why, or where, or how bad it hurts. This inability to determine when children with ASD are in pain could be attributed to the abnormal sensory processing previously described—which confuses caregivers, because it often contributes to a minimal (or lack of) response to seemingly painful events. Alternatively, it could be related to what parents look for in trying to determine whether their children are in pain.
Interestingly, a majority of the caregivers that stated their child “doesn’t feel pain,” “can handle pain more,” or “has a high tolerance for pain,” alluded to how they make this inference. They mentioned that their child “doesn’t cry,” “doesn’t say what is bothering him,” “is pain seeking,” “only cries a little,” “is nonverbal,” or “tells you in a calm manner that ‘it really hurts mommy’.” One needs to examine what these caregivers consider to be pain expressive behaviors in this population. If caregivers are expecting their children to cry, or respond dramatically to pain, they may wrongly assume that their children have a high tolerance for pain, simply because they do not display these more typical pain expressive behaviors.

Several caregivers acknowledged that their children experience pain differently, but they referred to this difference as being in the way they express their pain. They stated that they can determine when their child is in pain because “he tells me, but tends to be quite stoic,” “he will punch himself in the stomach if it hurts,” “he will lay around and sleep a lot,” “he will make up stories about getting away in a time machine,” or “because he never communicates that something hurts, you have to pay close attention to behavioral cues.” These caregivers have identified behaviors unique to their children that usually indicate they are in pain. One can see though, how some caregivers might interpret “he will punch himself in the stomach” as pain seeking, or having a high tolerance for pain—while other caregivers might identify that specific behavior as indicating pain in their particular child.

Some caregivers also reported that they have no trouble determining when their children are in pain because they are verbal, and can communicate when they are hurting. Comments regarding how they verbally communicate, however, indicate that those communication impairments typical of children with ASD permeate their ability to communicate pain. For example, they may “be quite stoic,” “just scream,” “just says several words and not express how
he feels about it at all,” “simply repeat a phrase he has heard,” or “reenact a story.” Children with reported high functioning autism will more often tell their caregivers when they are in pain. These findings warrant investigation into whether there are differences in pain expression between children with ASD of different diagnoses (i.e., Asperger’s disorder vs. PDD-NOS) and varying degrees of communicative ability.

Pain assessment in this population heavily relies on the perceptions and beliefs held by caregivers. This research aimed to illuminate some of the perceptions and beliefs that might be contributing to confusion regarding whether children with ASD feel pain. Clearly, the communication impairments described previously influence how children with ASD express pain. The theoretical model guiding this research proposes a relationship between “observer bias,” and an observer’s ability to “decode” pain expressive behaviors. If caregivers are watching for more typical pain expressive behaviors, their bias might cause them to inaccurately assume that their children are not in pain. This research demonstrates that there is a relationship between these two variables, and begins to illuminate what that relationship might be like.

These caregiver surveys indicated that while children with ASD might respond to painful events differently, caregivers do believe their children are capable of feeling pain. This conclusion is congruent with the findings reported by Nader et al. (2004) that “children with autism should be viewed as being as reactive to painful stimuli as children without autism and do not support beliefs about pain insensitivity in children with autism” (p. 96). Interestingly, these surveys indicate that sensory processing abnormalities in some children with ASD, and the perseverative nature of the disorder likely contribute to the confusion (especially experienced by caregivers) surrounding the pain experience in children with ASD. Astute caregivers have identified specific pain indicators (often non-typical pain indicators) that are uniquely exhibited
by their children. The second study conducted in this research aimed to investigate what autism experts believe about pain expression in ASD, and utilized the pain indicators identified by caregivers to further explore whether development of a pain assessment tool specific to this population might be feasible.

**Communication and Social Impairments in ASD and Pain Expression**

Findings from Study 1 and Study 2 further highlight how the communication and social impairments seen in ASD might influence pain expression. Deficits in joint attention are theorized to impair a child’s ability to develop social and communicative competencies. As previously described, joint attentional skills that children with ASD either lack (or have problems with) include referential looking, pointing and showing, following one’s gaze to look at an object with them, and visually following where others point. Tomasello’s (2003) theory of language acquisition describes how understanding the communicative intentions of others facilitates the emergence of language. The communication and social impairments in children with ASD affect the way they experience, and thus express, pain.

Study 1 findings emphasize that those children with greater communicative competency do outwardly express their pain. Caregiver comments that their children are often stoic when they describe their pain, or that they will verbally communicate their pain by repeating a phrase like “ants hurt you,” demonstrate that despite verbal abilities, communicative deficits in pragmatic language use and echolalia influence pain expression. Children with Asperger’s disorder might communicate their pain by using flat and emotionless speech, thus making it difficult to determine how severely they are hurting.

Study 2 findings highlight that the pain experience in children with ASD might be more of an *intrapersonal* experience, rather than an *interpersonal* experience. This is consistent with the core social deficits—also related to a lack of joint attention skills—seen in children with ASD.
Several expert-identified pain indicators that were unique to children with ASD seem to reflect an intrapersonal experience, for example: “rocking unusually,” “injuring oneself,” “banging his/her head,” “laying around, acting lethargic,” and “holding his/her head with both hands.” Several pain indicators helpful in assessing children with cognitive impairment and/or communicative impairments that were not considered relevant to children with ASD reflect an interpersonal experience. These include: “disinterest in surroundings,” and “less interaction with others, withdrawn.” These findings support the postulated significance of the social deficits seen in children with ASD to pain expression.

The Communications Model of Pain proposed by Hadjistavropoulos and Craig (2002) posits that social and communicative competencies influence the pain experience and pain expression. These research findings support the model’s proposed relationships, and preliminarily demonstrate that the communication and social impairments specifically seen in the ASD population appear to uniquely influence pain expression. When determining how to assess pain in ASD, it will be important to continue to test the proposed model, and consider how social and communicative features commonly seen in this population affect their ability to express pain. The studies conducted in this research established a foundation from which future studies may extend.

**Items Determined to be Relevant to Pain Assessment in ASD**

The second study in this research examined 77 potential pain indicators in children with ASD and aimed to determine which indicators are most relevant to pain assessment in this population. Expert review incorporated into instrument development ensures that items are added, removed, and revised, via an informed process. As previously mentioned, it is not uncommon for item lists utilized in instrument development to undergo several iterations of expert review, in order to ensure that the final set of items validly represent the construct of
interest. This section presents the Primary Pain Indicators identified as being most relevant to
pain assessment in this population. Additionally, the subset of items that were considered
relevant by a majority of the experts, however their calculated I-CVI fell below the pre-
established cut-off (the Secondary Pain Indicators) are discussed. The Secondary Pain Indicators
are evaluated, in light of this particular content validity process, and implications for leaving
these items in the list for another expert review are presented. Finally, comments and
suggestions provided by experts are reviewed.

**Primary Pain Indicators: Retained Items**

Fourteen items had a calculated I-CVI greater than .78, which indicated that they achieved
content validity for this construct of interest (pain). To review, these included: “rocking
unusually,” “lashing/striking out,” “being less active,” “banging his/her head,” “injuring
oneself,” “crankiness (being more cranky than usual),” “lying around/acting lethargic,”
“increased heart rate,” “furrowing brow,” “screaming/yelling/throwing tantrums,” “guarding
(protecting) or favoring a particular part of the body when moving,” “inconsolability (being
unable to console),” “tensing face,” and “grimacing.”

When these items are compared to items in pain assessment tools currently available for
use in related populations, there are similarities and differences. The Primary and Secondary
Pain Indicators derived from Study 2 are compared to three existing pain assessment tools in
Table 5-1. The existing assessment tools chosen for comparison are the Non-Communicating
Children’s Pain Checklist (NCCPC), the Pain Indicator for Communicatively Impaired Children
(PICIC), and a Pain Evaluation Scale developed for children with CP. These three assessment
tools were chosen for comparison because they have good psychometric properties and have
demonstrated validity in the populations for which they were developed. When this Investigator
generated the original item list, indicators from each of these existing tools were included so as to identify whether those indicators were relevant, or not relevant to children with ASD.

Significant differences imply that using existing pain assessment tools with individuals having an ASD would not be valid, and might lend to inaccurate pain assessment. For example, pain indicators identified by caregivers in Study 1 were added to the item list developed by this Investigator. Several of these items were subsequently validated by experts in Study 2, as being relevant to pain assessment in ASD (i.e., “rocking unusually”). These indicators however, are not seen in the existing pain assessment tools available for use in similar and related populations. Furthermore, several of the indicators that are utilized to assess pain in cognitively impaired, non-communicating children were not considered relevant to pain assessment in ASD. These included, for example: “disinterest in surroundings,” “less interaction with others, withdrawn,” flinching or moving away, sensitive to touch,” and “jumping around, agitated, fidgety.” Given the underlying nature of ASD, these behaviors are commonly exhibited due to their disorder (i.e., “less interaction with others, withdrawn”), and thus would not be useful in pain assessment.

Other pain expressive indicators included in existing assessment tools, but not determined to be relevant to pain assessment in ASD, involved behaviors more applicable to children with CP—because of their associated motor impairments—such as “stiff, spastic, tense and rigid,” “floppy,” or “searches for an unusual position that calms.” Additionally, several physiological indicators and changes in daily activity that were included in other assessment scales were not considered relevant to pain assessment in ASD (“shivering,” “sweating, perspiring,” “eating less, not interested in food,” and “increase in sleep”).

A number of pain indicators identified by caregivers were subsequently determined to be relevant to pain assessment in ASD by pain and autism experts. These indicators included
“rocking unusually,” “lashing/striking out,” “injuring oneself,” and “banging his/her head.”

Other unique changes likely to indicate pain in this population, according to Study 2, include “increased heart rate,” and “lying around/acting lethargic.” One other finding that consistently arose throughout this research is that while there may be some unique pain indicators applicable to most children with ASD, there will also be those pain indicators identified by caregivers that are completely unique to the one child. Despite attempts to identify common pain indicators in children with ASD, pain assessment in this population also seems to rely heavily on “deviations from normal.”

**Secondary Pain Indicators: Item Retention and Revision**

Several experts explained how they conceptualized whether an item was “relevant” to pain assessment in ASD. They indicated that they determined an item’s relevance based on whether a particular indicator applied to all children with ASD (thus rating it as “highly relevant”), most children with ASD (“quite relevant”) some children with ASD (“somewhat relevant”) or not occurring at all in children with ASD (“not relevant”). As a result, a number of items (n = 19) were rated “somewhat relevant” by experts, but because the I-CVI is calculated using only those ratings of “quite” and “highly” relevant, these items fell below the pre-established I-CVI cut-off of .78. Given that these items were close to the desired I-CVI of .78, and according to recommendations by Polit, Beck, and Owen (2007), it was determined that they should be closely examined before determining whether they should be discarded. Table 5-2 lists the items with a calculated I-CVI = .67. This I-CVI (.67) was obtained when four of six experts rated the item as “quite” or “highly” relevant, and the other two rated the item as either “not” or “somewhat” relevant.

When listing each item with an I-CVI of .67, as presented in Table 5-2, the differences in how experts rated each of these items can be more closely examined. Conceptually, there is a
difference between the item where ALL experts believed it was “somewhat relevant” or higher, and the item where two experts believed it was “not relevant” at all, and the other four believed it was “quite” or “highly” relevant. These two items however, given the way the I-CVI is calculated, would have an identical I-CVI. This is clearer when one looks at a calculated “sum” score for each item. If 1 = “not relevant,” 2 = “somewhat relevant,” 3 = “quite relevant,” and 4 = “quite relevant,” a summed score can be generated for each item, depending on how relevant each expert rated the item. The items can then be rank-ordered and one can look at the relationships between them.

Despite all items having and I-CVI of .67, the variability becomes evident when one looks at the range of sum scores (15–19) amongst all items with an I-CVI or .67. For example, experts rated “sleeping less” as “not relevant” (n = 1), “somewhat relevant” (n = 1), and “quite relevant” (n = 4). The sum score for this item is 15 (1+2+3+3+3+3 = 15). “Stating/Repeating a specific pain word” was rated “somewhat relevant” (n = 2), “quite relevant” (n = 1), and “highly relevant” (n = 3). The sum score for this item is 19 (2+2+3+4+4+4 = 19). There is a difference between the item that has one expert rating it “not relevant” and does not have any experts rating it as “highly relevant,” and the item that was never rated “not relevant” and has three experts rating it “highly relevant.” The methodology used to generate the I-CVI does not take this variability amongst the items with the same I-CVI into consideration, and by dichotomizing the 4-point Likert scale, information about each item is lost, and might be to the detriment of the scale’s development. A number of items that might be relevant to pain assessment in ASD, which is a highly heterogeneous population, would be removed if the strict .78 cut-off is used, without examining those items just below the cut-off.
Several of the Secondary Pain Indicators also need to be revised, as suggested by the expert panel. For example, “crying” should be combined with “moaning, whining, whimpering,” to generate one pain expressive behavior that captures what the two separate indicators are reflecting: different degrees of “vocalization.” So the new item, once the response format is determined, might be “Vocalization.” Caregivers would then rate their children’s vocalization on a Likert scale, choosing either: “0” (quiet), “1” (soft vocalization like moaning, whining, or groaning), “2” (crying), or “3” (screaming, yelling). In doing this, the indicator “screaming/yelling/throwing tantrums” would also be revised, and subsequently separated into “screaming/yelling” and “throwing tantrums.” “Throwing tantrums” would be taken out, resulting in a new item’s creation—it was mentioned by one expert that throwing a tantrum seems different than perhaps screaming or yelling in pain.

Whenever items are revised, it is suggested that they remain in the item list and undergo and second expert review to determine if revision made them more relevant to the instrument’s construct of interest—in this case, assessing pain in children with ASD. All items that underwent some type of revision were retained in the item list for future examination of the items to further establish the content validity of a final set of items.

The question arises in this research, what should be done with the Secondary Pain Indicators? Given a) the early stage of this instrument’s development, b) the methodological challenges associated with dichotomizing the I-CVI, c) challenges associated with assessing pain in such a heterogeneous population, and d) the fact that this initial iteration revealed a need for several Secondary Item revisions, it was determined that the Primary and Secondary Pain Indicators would both be utilized to generate the next item list, and that an additional expert review is likely warranted. Items subsequently not meeting Lynn’s (1986) criteria for adequate
content validity will be removed after the next review by experts, and a final set of items will include only those indicators deemed most relevant to pain assessment in ASD by experts.

The findings from Study 1 highlight the variability seen in children with ASD as reported by caregivers. Children with ASD present with various patterns of strengths and weakness, and thus it may be impractical to develop one assessment tool appropriate for all children with ASD. The Primary Pain Indicators highly endorsed by experts indicate that there may be several pain expressive behaviors unique to this population and somewhat common across all children with ASD. The Secondary Pain Indicators might be relevant to subpopulations of children with ASD, or simply not all children with ASD. Experts believed them to be “somewhat relevant” but simply not “highly relevant.” These items need to be further explored, systematically, in another round with experts—or perhaps in a subsequent study where the assessment tool is actually utilized by caregivers and providers of children with ASD.

**Items Removed from the Item List Following Expert Review.**

All items with an I-CVI less than .67 (n = 44) were removed. Least relevant items included “laughing,” “humming/singing,” “becoming floppy, or limp,” “interacting more with others,” “eating more than usual,” and “chewing, moving tongue around and/or out, smacking lips.” A number of items may be relevant to pain assessment in individual children with ASD, or perhaps in small subgroups of children with ASD. These items not commonly seen in ASD reflect a more individualized pain process. The next section discusses how to accommodate these individually significant (but less common) pain indicators, in light of several expert comments.

**Expert Comments and Suggestions**

The Investigator solicited experts who could address the relevance of items to pain assessment in ASD and offer insight regarding the overall development of this pain assessment
tool. Grant and Davis (1997) suggest using experts to help ensure items are clear, concise, and comprehensive. Expert suggestions regarding item revision were considered, and input on how to categorize the resulting pain indicators in an assessment tool was well received by this investigator. DeVellis (2003) points out however, that the “final decision to accept or reject the advice of your experts is your responsibility as the scale developer” (p. 87). Given that suggestion, expert comments are presented here, but are considered in light of the overall goals and plans for this instrument’s development.

A Need for Individualized Pain Assessment in this Population.

Contrary to what experts believed to be relevant to pain assessment, several caregivers reported in Study 1 that “laughing” was sometimes an indication of pain in their children. This discord speaks to the fact that there will likely continue to be unique and individual pain expressive indictors exhibited by children in this population. Expert suggestions that this instrument include a section focused on development of an “individual pain profile” for use in children with ASD were considered. This is similar to how Solodiuk and Curley (2003), as previously described, approach pain assessment in nonverbal children with severe cognitive impairment. Their instrument provides examples of pain expressive behaviors commonly seen in nonverbal children with cognitive impairment, and then asks caregivers to work from the suggested list to generate a pain expression profile unique to their child at various intensities of pain.

Given the heterogeneity seen in children with ASD, it may be helpful to incorporate some of what Solodiuk and Curley (2003) have found useful. Perhaps this assessment tool could be comprised of two parts. An initial part (Part A) might recommend that caregivers watch for particular (those most common) pain expressive behaviors. This would be especially helpful for newly diagnosed children, or to caregivers who are watching for more typical pain expressive
indicators—and thus concluding that their children do not feel pain. A second part (Part B) will allow caregivers to begin to determine their child’s own pain profile. Once a comprehensive picture of how the child expresses physical pain is established, this tool can be distributed to other individuals involved in his or her care. In this way, the two methods of pain assessment are merged. Suggestions of what to watch for are offered, however it is also acknowledged that each child is different and unique in his or her pain expression.

The Role of Direct Behavioral Observation in Instrument Development

The original item list generated for Study 2 aimed to include a wide variety of possible pain indicators in children with ASD. When the items were reviewed by experts, the role of direct observation in the instrument’s development, and subsequent use with children was questioned and is therefore addressed here. It was acknowledged that several of the pain expressive indicators under review by experts are not directly observable, and require one to rely on caregiver reports of “deviation from normal.” These items include: “unhappiness,” “acting ‘off’ according to caregiver,” and “crankiness.” Two were considered highly relevant to pain assessment in this population, and therefore clarification regarding the definition and operationalization of these items is necessary before re-evaluating the item list.

This research illuminated the heavy emphasis on deviations from normal when assessing whether this population of children is in pain. While there were several objective, observable, and measurable indicators presumed to be relevant to pain assessment in ASD (i.e., “furrowed brow,” “banging his/her head,” “injuring oneself,” “grimacing,” “guarding,” and “increased heart rate”), many other identified indicators were subjective, and require a) knowledge of the child’s baseline, and b) monitoring for changes from normal (i.e., “crankiness,” “being less active,” “rocking unusually,” “acting ‘off’,” “irritability”). Given this, there might need to be sections of the pain assessment tool that are objective and quantifiable—these could be used by someone
unfamiliar with the child—while other sections of the final assessment tool might be based on caregiver judgments regarding what is normal in their child. Caregiver information regarding their child’s baseline, and thus deviations from normal will be critical to comprehensive pain assessment in this population. Future research into whether caregivers’ subjective information correlates with objective, measurable indicators is needed. In this way, direct observation of children would be extremely valuable.

When beginning to explore pain assessment in children with ASD, this Investigator considered study designs that incorporated direct observation of children presumed to be in pain, in order to inductively generate a list of observable behaviors potentially relevant to pain assessment in this population. The confusion that surrounded pain assessment in children with ASD however, made it difficult to theoretically determine what to “watch for” when observing this population. The reports that these children do not react to pain in the same manner as typically developing children highlighted the need for initial pilot work aimed to simply characterize the issues encountered when assessing pain in this population. Thus the studies conducted in this research gathered descriptive information in order to deductively determine what types of pain expressive behaviors one would “watch for” when directly observing children. The next steps in this instrument’s development will certainly incorporate direct observation, and attempt to correlate caregiver reports with certain pain expressive behaviors that are directly observable to someone unfamiliar with the child. Direct observation without any informed information regarding how children with ASD express pain would have been limited in its usefulness.

Next Steps in this Instrument’s Development

This instrument development process began by attempting to articulate a) if children with ASD experience and express pain differently than typically developing children and b) whether it
is possible to identify a set of pain expressive behaviors unique to this population. Study 1 findings indicate that children with ASD are capable of experiencing and expressing pain, however their pain expression is unique. Study 2 validates this uniqueness by identifying pain indicators specific to children with ASD, and by demonstrating that indicators relevant to other, similar, populations of children are not considered to be relevant to this population. Following Study 2, the resulting list of pain expressive behaviors deemed relevant to children in ASD needs to be further examined. Therefore, the next steps in this instrument’s development involve a) the revision of particular items, b) re-evaluation of the new set of items (having been revised and re-categorized), c) determination of how each item should be scaled, and d) additional testing of the final set of items to determine whether the resulting pain assessment tool is valid and reliable in children with ASD. In formalizing the final set of items into a scale, each pain indicator will need to have its operational definition further articulated.

Future item analysis involves examining the final instrument’s applicability to the population of interest. It will need to be determined whether caregivers can utilize the tool to discern pain in their children with ASD. The purpose of follow up research will be to a) examine how the items relate to one another, and how each item influences the overall statistics of the tool (internal consistency), b) conduct correlation analyses to examine whether total-item (summed) scores are different for children with various characteristics within the ASD population, c) examine the endorsement frequency—the proportion of caregivers who give each response option to an item—of each item, and d) analyze the caregivers’ mean usefulness ratings. It will be helpful to know which items caregivers endorse, and how useful the items are in determining whether their children are in pain.
The next step following item analysis and adjustment will be to further test the reliability and validity of the existing tool. Establishing construct validity will require a series of studies. Each additional study demonstrating that the instrument is useful further validates it as a pain assessment tool for the specified population. The purpose of a validation study is to determine whether the tool maintains adequate internal consistency, to establish intrarater reliability, to examine concurrent and discriminant validity, and to determine whether caregivers can use it to discriminate between episodes of pain and nonpain. The most significant analysis for this particular study design will be to determine if the scores on the assessment tool differ between pain and nonpain episodes (discriminant validity). This repeated measures design requires Repeated Measures Analysis of Variance (RANOVA). The same caregivers will fill out the checklist during two separate pain events, and therefore it will possible to examine the tool’s stability over time. It can be determined if the number of items observed is consistent, and whether the same items are endorsed for a child during two separate pain events. Mixed design RANOVA will allow one to examine whether there are differences between groups of subjects regarding a specific factor (i.e., communication ability, age, etc). Finally, because the same caregivers will utilize the tool on two different occasions, it will be possible to examine intrarater reliability in terms of caregiver use of the tool. Kappa coefficients analyzing the caregiver’s agreements when utilizing the tool at each pain event will establish intrarater reliability. Streiner and Norman (1995) propose that acceptable reliabilities reported for instruments need to be greater than .75.

Future studies will also incorporate video observation of children with ASD experiencing pain in perhaps a more clinical setting (post-operative pain for example) to establish other required forms of reliability (interrater) between different observers (observer, caregiver, staff
nurse, researcher). Additionally, correlating instrument scores with perhaps facial coding could help further establish concurrent validity—given that facial coding is a well-established valid method for assessing for pain in children as somewhat of a “gold standard.” There are no behavioral observation protocols well validated for assessing pain in children with ASD, although there are specific behaviors that have been validated as observable pain indicators in other populations (elders with dementia, for example). Capturing pain behavior on video would allow for additional exploration into how children with ASD exhibit pain from the perspective of pain experts, rather than caregiver report. This will be useful if the caregivers have difficulty discerning pain in their children with ASD, even after being given a repertoire of behaviors thought to indicate pain in this population.

Limitations

Given the paucity of information available to begin to investigate the general question approached in this research, these studies were preliminary in nature, and aimed to explore and describe issues important to assessing pain in children with ASD. Several limitations were identified as each study was conducted, and these include the survey response rate, and the quasi-qualitative design utilized in Study 1.

Survey Response Rate

A total of 400 surveys were mailed to caregivers of children with ASD in Study 1. Eighty-eight surveys were completed and returned by mail, resulting in a 22% return rate. This lower response rate is problematic because it “influences the validity of inferring that the sample represents the population” (Hulley, Newman, & Cummings, 2001, p. 31). However, a lower response rate might be expected in this particular population given the tremendous challenges associated with caring for a child with this disorder. Many of these caregivers are overwhelmed by the responsibilities required to ensure that their children receive the best intervention and care.
possible. A better response rate might have been achieved if an incentive was included in the study design. Of those caregivers who did respond, almost all included written descriptive responses, and 80% provided their name and number for a follow up call. This willingness to provide detailed information highlighted the strong desire to discuss and share concerns regarding pain assessment in their children with others.

**Quasi-Qualitative Design**

Study 1 afforded caregivers the opportunity to provide written descriptions of their thoughts regarding the pain experience in their children. These written responses were examined by the investigator and an autism expert and commonly reported issues were tallied. The most frequently reported issues were utilized to identify a few themes amongst this descriptive data. If the focus of this research was to specifically describe how caregivers perceived the pain experience in their children, a purely qualitative study would have been informing. However, the goal of Study 1 was to simply establish if pain assessment in children with ASD was a problem. Nothing in the literature to date addressed whether pain assessment in ASD warrants an investigation. Study 1 was originally designed to obtain a snapshot of whether caregivers and healthcare providers would benefit from research focused on pain assessment in ASD. The sheer volume of written descriptive information provided by caregivers was surprising. Given this volume of descriptive data, more rigorous and formal qualitative inquiry might have been even more revealing.

Other studies attempting to develop pain assessment tools for unique populations utilize caregivers to draft an initial item list, as done for this research. Some common methods used to solicit caregiver perspectives include having them participate in focus groups, fill out checklists, and answer survey questions. This study was designed in a similar fashion, foregoing the option
to conduct an in-depth qualitative study, and choosing instead to complete two preliminary studies aimed at facilitating instrument development for this research.

**Nursing Implications and Clinical Significance**

As members of dynamic and interdisciplinary health care teams providing patient care, nurses are often directly responsible for assessing pain and initiating means to relieve patient suffering. In the absence of self-report, it is often the astute assessment of the subtleties in a patient that allow a good nurse to identify, intervene, and relieve pain. This awareness is even more critical when assessing and managing pain in several vulnerable populations of children. As the need for better pediatric pain assessment emerged in the 90s, Ferrell, McCaffery, & Ropchan (1992) surveyed clinical nurses and identified barriers to adequate pain management. Top barriers included *lack of knowledge* on the part of the physicians, nurses, patient and family members, and *system issues* involving administration of pain medication. An effort has been made over the past several decades to increase nursing knowledge of pain assessment and management in children, and to prioritize pain assessment and management in the clinical setting.

This nursing research aimed to equip parents and healthcare providers with the additional knowledge and understanding required to determine if and when a child with ASD might be experiencing pain. In pursuing the answers to the proposed research questions, it was the hope that many of the myths and controversial issues surrounding the question of whether children with ASD are capable of feeling pain can be eliminated. Not only are nurses directly involved in patient care management, but nurses cross a myriad of settings and function at various practice levels. Dissemination of these research findings will equip nurses at the bedside, and nurses in the clinic setting, with the ability to better assess pain in children with ASD. Furthermore, the perceptions that children with ASD are incapable of feeling pain can be corrected. Nurses
function as clinicians, advocates, and educators. An accurate understanding of how children with ASD experience pain will ensure that parents of newly diagnosed children, or parents having a difficult time determining when their children with ASD are in pain, are supported and taught about pain assessment and management in their children.

**Future Directions**

Instrument development requires diligence and dedication. A series of studies are required to ensure that a valid and reliable means of assessing the construct of interest results. The studies conducted in this research lay a foundation from which future instrument development can build. Future research aims to continue refining pain expressive indicators specific to children with ASD. As these indicators are further articulated, additional investigation into the variability in pain expression found along the spectrum is warranted. It is likely that there are subgroups of children with ASD who have particular features influencing their specific pain expressive abilities (i.e., those with severe abnormal sensory processing).

The Communications Model of Pain (Hadjistravopoulos & Craig, 2002) highlights a variety of relationships that need to be explored. This research demonstrated that caregiver perceptions, and existing myths or biases, likely influence one’s ability to decode pain expressive behaviors in children with ASD. By focusing on the nonverbal and involuntary mechanisms of pain expression, it is possible to determine how children with ASD behaviorally exhibit their physical discomfort. Additional investigation into how children with ASD respond to different *types* of pain is still needed. It is possible that acute pain elicits a more reflexive, involuntary response, while everyday bumps and bruises (or more chronic types of pain) require a greater understanding of the social context surrounding the pain experience. This requirement (understanding the social context) would be impaired in a population of children with social and communicative impairments (i.e., ASD).
Additionally, future work will need to focus on caregivers’ abilities to accurately decode their child’s pain expressive behavior, and examine whether their knowledge is appreciated and utilized by healthcare providers across settings. Complaints that healthcare providers dismiss caregiver beliefs about their children’s pain are alarming, and speak to a lack of partnership between some healthcare providers and caregivers in this population.

Finally, work to disseminate findings following the ultimate development of a pain assessment tool available for use in children with ASD will include examining the applicability of this tool to children in a variety of settings. Hopefully, illuminating the pain assessment issues specific to children with ASD will debunk circulating myths, and allow everyone involved in the care of a child with ASD to adequately assess, treat, and manage their pain.
<table>
<thead>
<tr>
<th>Study 2: Items identified as relevant to pain assessment in ASD by experts</th>
<th>Non-Communicating Children’s Pain Checklist (NCCPC); 30 Items</th>
<th>Pain Indicator for Communicatively Impaired Children (PICIC); 6 Items</th>
<th>Pain Evaluation Scale for CP; 10 Items</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Primary pain indicators (I-CVI &gt; .78)</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Rocking unusually</td>
<td>—</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td>Lashing/striking out</td>
<td>—</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td>Guarding (protecting) or favoring a part of the body when moving</td>
<td>Protecting, favoring, guarding part of body that hurts</td>
<td>—</td>
<td>Coordinated defensive reaction of an area supposed painful protection of painful areas</td>
</tr>
<tr>
<td>Increased heart rate</td>
<td>—</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td>Tensing face</td>
<td>—</td>
<td>Screwed up or distressed looking face</td>
<td>Painful expression</td>
</tr>
<tr>
<td>Furrowing brow</td>
<td>Furrowed brow</td>
<td>Screwed up or distressed looking face</td>
<td>Painful expression</td>
</tr>
<tr>
<td>Grimacing</td>
<td>—</td>
<td>Screwed up or distressed looking face</td>
<td>Painful expression</td>
</tr>
<tr>
<td>Screaming/yelling/throwing tantrums</td>
<td>Screaming/yelling</td>
<td>Screaming, yelling, groaning or moaning</td>
<td>—</td>
</tr>
<tr>
<td>Injuring oneself</td>
<td>—</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td>Banging his/her head</td>
<td>—</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td></td>
<td>—</td>
<td>—</td>
<td>Disinterest in surroundings</td>
</tr>
<tr>
<td>Lying around/acting lethargic</td>
<td>—</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td>Being less active</td>
<td>Not moving, less active, Quiet</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td>Crankiness (being more cranky than usual)</td>
<td>Not cooperating, cranky, irritable, unhappy</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td>Study 2: Items identified as relevant to pain assessment in ASD by experts</td>
<td>Non-Communicating Children’s Pain Checklist (NCCPC)</td>
<td>Pain Indicator for Communicatively Impaired Children (PICIC)</td>
<td>Pain Evaluation Scale for CP</td>
</tr>
<tr>
<td>---</td>
<td>---</td>
<td>---</td>
<td>---</td>
</tr>
<tr>
<td><strong>Inconsolability (unable to console)</strong></td>
<td>Difficult to distract, notable to satisfy or pacify</td>
<td>Difficult to comfort or console</td>
<td>—</td>
</tr>
<tr>
<td><strong>Secondary pain indicators (I-CVI = .67)</strong></td>
<td>Less interaction with others, withdrawn</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td>Acting “off” according to caregiver: not acting like him/her self</td>
<td>Flinching or moving body part away, sensitive to touch</td>
<td>Flinches or moves away if touched</td>
<td>—</td>
</tr>
<tr>
<td>Body appears stiff or tense</td>
<td>Tonic troubles: Stiffness</td>
<td>—</td>
<td></td>
</tr>
<tr>
<td>Stiff, spastic, tense, rigid Floppy</td>
<td>—</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td>Crying (with or without tears)</td>
<td>Crying (moderately loud)</td>
<td>Crying (with or without tears)</td>
<td>Crying</td>
</tr>
<tr>
<td>Moaning, whining, whimpering (softly)</td>
<td>Moaning, whining Whimpering</td>
<td>—</td>
<td>Moaning or inaudible cries</td>
</tr>
<tr>
<td>Stating/repeating a specific pain-related word (i.e., hurt, ouch)</td>
<td>Exhibits a specific word/sound for pain (or type of laugh)</td>
<td>—</td>
<td>Does not communicate with others</td>
</tr>
<tr>
<td>Moving body in a specific way to show pain</td>
<td>—</td>
<td>Searches for an unusual position that calms</td>
<td>—</td>
</tr>
<tr>
<td>Seeking comfort or physical touch</td>
<td>Seeking comfort or physical closeness</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td>Touching, rubbing, picking, and/or tapping a specific part of the body</td>
<td>Gesturing to or touching part of body that hurts Shivering</td>
<td>—</td>
<td>—</td>
</tr>
</tbody>
</table>
Table 5-1. Continued

<table>
<thead>
<tr>
<th>Study 2: Items identified as relevant to pain assessment in ASD by experts</th>
<th>Non-Communicating Children’s Pain Checklist (NCCPC)</th>
<th>Pain Indicator for Communicatively Impaired Children (PICIC)</th>
<th>Pain Evaluation Scale for CP</th>
</tr>
</thead>
<tbody>
<tr>
<td>Changing color, (flushing/redness)</td>
<td>Changing in color, (pallor)</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td>Producing tears (without overt crying or vocalizations)</td>
<td>Perspiring</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td>Increased rate of breathing</td>
<td>Sweating, perspiring</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td></td>
<td>Change in color, (pallor)</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td></td>
<td>Gasping</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td></td>
<td>Breath holding</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td>Sleeping less</td>
<td>Eating less, not interested in food</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td>Grinding teeth, clenching jaw down</td>
<td>Increase in sleep</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td>Frowning, turning down mouth</td>
<td>Increase in spontaneous movement (voluntary/not)</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td>Pouting, quivering, puckering lips</td>
<td>Jumping around, agitated, Fidgety</td>
<td>—</td>
<td>—</td>
</tr>
</tbody>
</table>

Note: Primary and Secondary Pain Indicators are compared to items included in existing pain assessment tools for use in related, but different, populations of children. Italicized items indicate that items are either unique to children with ASD or not relevant to children with ASD (because they were not validated in Study 2 by autism experts for use in this population).
Table 5-2. Study 2: Variability in expert-rated relevance amongst all items with an I-CVI = .67

<table>
<thead>
<tr>
<th>Pain behavior/indicator</th>
<th>Not relevant</th>
<th>Somewhat relevant</th>
<th>Quite relevant</th>
<th>Highly relevant</th>
<th>Item - CVI</th>
<th>Sum</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sleeping less</td>
<td>1</td>
<td>1</td>
<td>4</td>
<td>—</td>
<td>.67</td>
<td>15</td>
</tr>
<tr>
<td>Irritability (being more irritable than usual)</td>
<td>—</td>
<td>2</td>
<td>4</td>
<td>—</td>
<td>.67</td>
<td>16</td>
</tr>
<tr>
<td>Seeking comfort/physical touch</td>
<td>—</td>
<td>2</td>
<td>4</td>
<td>—</td>
<td>.67</td>
<td>16</td>
</tr>
<tr>
<td>Becoming more aggressive (hitting, pinching, biting others)</td>
<td>—</td>
<td>2</td>
<td>4</td>
<td>—</td>
<td>.67</td>
<td>16</td>
</tr>
<tr>
<td>Acting angry</td>
<td>—</td>
<td>2</td>
<td>4</td>
<td>—</td>
<td>.67</td>
<td>16</td>
</tr>
<tr>
<td>Widening eyes</td>
<td>1</td>
<td>1</td>
<td>3</td>
<td>1</td>
<td>.67</td>
<td>16</td>
</tr>
<tr>
<td>Frowning, turning down mouth</td>
<td>1</td>
<td>1</td>
<td>3</td>
<td>1</td>
<td>.67</td>
<td>16</td>
</tr>
<tr>
<td>Pouting, quivering, puckering lips</td>
<td>1</td>
<td>1</td>
<td>3</td>
<td>1</td>
<td>.67</td>
<td>16</td>
</tr>
<tr>
<td>Producing tears (without overt crying or vocalizations)</td>
<td>—</td>
<td>2</td>
<td>3</td>
<td>1</td>
<td>.67</td>
<td>17</td>
</tr>
<tr>
<td>Increased rate of breathing</td>
<td>—</td>
<td>2</td>
<td>3</td>
<td>1</td>
<td>.67</td>
<td>17</td>
</tr>
<tr>
<td>Acting “off” according to caregiver: not acting like him/herself</td>
<td>—</td>
<td>2</td>
<td>3</td>
<td>1</td>
<td>.67</td>
<td>17</td>
</tr>
<tr>
<td>Grinding teeth, clenching jaw down</td>
<td>—</td>
<td>2</td>
<td>3</td>
<td>1</td>
<td>.67</td>
<td>17</td>
</tr>
<tr>
<td>Squinching, squinting eyes</td>
<td>—</td>
<td>3</td>
<td>1</td>
<td>2</td>
<td>.67</td>
<td>17</td>
</tr>
<tr>
<td>Crying (with or without tears)</td>
<td>—</td>
<td>2</td>
<td>2</td>
<td>2</td>
<td>.67</td>
<td>18</td>
</tr>
<tr>
<td>Moaning, whining, whimpering (softly)</td>
<td>—</td>
<td>2</td>
<td>2</td>
<td>2</td>
<td>.67</td>
<td>18</td>
</tr>
<tr>
<td>Changing color (flushing, redness)</td>
<td>—</td>
<td>2</td>
<td>2</td>
<td>2</td>
<td>.67</td>
<td>18</td>
</tr>
<tr>
<td>Touching, rubbing, picking, and/or tapping a specific part of the body</td>
<td>—</td>
<td>2</td>
<td>2</td>
<td>2</td>
<td>.67</td>
<td>18</td>
</tr>
<tr>
<td>Holding his/her head with both hands for prolonged periods of time</td>
<td>—</td>
<td>2</td>
<td>2</td>
<td>2</td>
<td>.67</td>
<td>18</td>
</tr>
<tr>
<td>Stating/Repeating a specific pain-related word (i.e., hurt, ouch)</td>
<td>—</td>
<td>2</td>
<td>1</td>
<td>3</td>
<td>.67</td>
<td>19</td>
</tr>
</tbody>
</table>

Note: The numbers in each column indicate how many experts rated the item as “not,” “somewhat,” “quite,” or “highly” relevant. The differences in sum score highlight the variability amongst all items with the same calculated I-CVI (.67).
Dear Parent or Guardian,

I am a nursing student at the University of Florida, and I am conducting a research study exploring how children with autism experience and express pain. The purpose of the study is to understand the thoughts parents have about whether their child feels, and can communicate when he or she is in pain.

Participation in this study involves completing the attached brief survey. It should only take about 5-10 minutes to answer the three questions. Your responses will be kept completely anonymous. Please do not write your name on the survey. All responses will be grouped together, and there will be no way to link your responses to your identity. The completed surveys can be placed in the sealed envelope and either dropped in the box located by the exit, or may be returned to the staff member who gave it to you. Sealing your survey in the envelope ensures that your responses will not be opened by anyone other than myself.

Participation in this study is completely voluntary. You do not have to fill out the survey. You may withdraw from the study at any time. If you choose not to participate, this will have no effect on the care or treatment of your child or family. Only the investigators involved in this study have the legal right to review your responses, and I assure you that we will protect the confidentiality of your information. If results from the study are published, anonymity will be maintained. If you have questions about your rights as a research participant, you may call the Institutional Review Board at 352-846-1494.

I will not be able to pay you for participating, but it will not cost you anything to participate.

Thank you for you time in reading this letter and thinking about completing the survey. If you have any questions, please call Melissa Dodd (352-392-9409), or ask the staff presenting you with this survey.

Sincerely,

Melissa Dodd, MSN, ARNP
Doctoral Student
College of Nursing, University of Florida
APPENDIX B
STUDY 2: ITEM LIST FOR EXPERT REVIEW

Instructions:

Please review each item and determine whether it might be relevant for assessing pain in children with ASD (especially nonverbal children). Recall that these items will undergo further testing/validation, and do not reflect the final version of the pain assessment scale. You can simply place an “x” under the appropriate column below, re-save the completed document with your name, and return to me via email.

It is the goal of this process to obtain a number of items that might be relevant to this population by weeding out irrelevant indicators. If there are behaviors that need to be considered but are not on this list, feel free to suggest that they be added. Those items rated as relevant by the majority of expert reviewers will remain in the item pool for the next step in the development of this tool. The final list of items will be utilized to construct a more formal pain assessment tool which will undergo additional expert review for item style (wording, clarity), and then be distributed to a test sample to evaluate overall tool comprehensiveness (understanding instructions, response format, length, etc).

<table>
<thead>
<tr>
<th>Pain Behavior /Indicator</th>
<th>NOT Relevant</th>
<th>SOMEWHAT Relevant</th>
<th>QUITE Relevant</th>
<th>HIGHLY Relevant</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Vocal/Verbal Indicators</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Crying (with or without tears)</td>
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<tr>
<td>Screaming/Yelling/Throwing Tantrums</td>
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<tr>
<td>Stating/Repeating a specific pain-related word (i.e. hurt, ouch)</td>
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<tr>
<td>Stating/Repeating a specific non-pain word or phrase (i.e. bat, toe…)</td>
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<tr>
<td>Babbling (Saying something incomprehensible)</td>
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<tr>
<td>Moaning, whining, whimpering (softly)</td>
<td></td>
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<tr>
<td>Humming/Singing</td>
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<tr>
<td>Laughing</td>
<td></td>
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<tr>
<td>Vocalizing less than normal, getting quiet</td>
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<tr>
<td><strong>Physiological</strong></td>
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<tr>
<td>Sweating, perspiring</td>
<td></td>
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<tr>
<td>Producing tears (without overt crying or vocalizations)</td>
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<tr>
<td>Shivering, shaking</td>
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<tr>
<td>Increased rate of breathing</td>
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<tr>
<td>Decreased rate of breathing</td>
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<tr>
<td>Changing color (paleness)</td>
<td></td>
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<tr>
<td><strong>Activity/Movement</strong></td>
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<tr>
<td>Sitting still</td>
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<tr>
<td>Fidgeting, becoming more active,</td>
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<tr>
<td>jumping around (motor restlessness)</td>
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<tr>
<td>Muscle tensing, rigidity</td>
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<tr>
<td>Guarding (protection) or favoring a</td>
<td></td>
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<tr>
<td>particular part of the body when</td>
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<tr>
<td>moving</td>
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<tr>
<td>Touching, rubbing, picking, and/or</td>
<td></td>
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</tr>
<tr>
<td>tapping a specific part of the body</td>
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<tr>
<td>Seeming hypersensitive (overly</td>
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<td></td>
<td></td>
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<tr>
<td>sensitive) to touch</td>
<td></td>
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<tr>
<td>Seeming less sensitive to touch</td>
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<tr>
<td>Becoming floppy, or “goes limp”</td>
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<tr>
<td>Being less active</td>
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<tr>
<td>Lashing/Striking out</td>
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<tr>
<td>Changing his/her usual body language</td>
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<tr>
<td>Holding his/her head with both hands</td>
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<tr>
<td>for prolonged periods of time</td>
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<td></td>
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<tr>
<td>Stomping foot</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Clapping hands loudly</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Rocking unusually</td>
<td></td>
<td></td>
<td></td>
<td></td>
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<tr>
<td>Scratching a specific part of the body</td>
<td></td>
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<td></td>
<td></td>
</tr>
<tr>
<td>Banging his/her head</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Clapping hands loudly</td>
<td></td>
<td></td>
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<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th><strong>Behavioral</strong></th>
</tr>
</thead>
<tbody>
<tr>
<td>Holding breath</td>
</tr>
<tr>
<td>Injuring oneself</td>
</tr>
<tr>
<td>Crankiness (being more cranky than</td>
</tr>
<tr>
<td>usual)</td>
</tr>
<tr>
<td>Cooperating less than normal</td>
</tr>
<tr>
<td>Irritability (being more irritable</td>
</tr>
<tr>
<td>than usual)</td>
</tr>
<tr>
<td>Unhappiness</td>
</tr>
<tr>
<td>Interacting less than normal with</td>
</tr>
<tr>
<td>others</td>
</tr>
<tr>
<td>Interacting more than normal with</td>
</tr>
<tr>
<td>others</td>
</tr>
<tr>
<td>Seeking comfort/physical touch</td>
</tr>
<tr>
<td>Isolating oneself more, hiding</td>
</tr>
<tr>
<td>Moving to, or staying in, a specific</td>
</tr>
<tr>
<td>location or “safe” space</td>
</tr>
</tbody>
</table>

- Changing color (flushing, redness)
- Increased heart rate
- Fever
- Feeling cool or cold to touch
| Perseverating more on a particular item |  |  |  |
| Making up stories or “reenacting” a painful circumstance |  |  |  |
| Acting “off” according to caregiver: not acting like him/herself |  |  |  |
| Becoming more aggressive (hitting, pinching, biting others) |  |  |  |
| Inconsolability (being unable to console) |  |  |  |
| Loosing interest in things that are usually extremely important or interesting to child |  |  |  |
| Increasing number and/or frequency of repetitive behaviors |  |  |  |
| Acting angry |  |  |  |
| Decreasing number and/or frequency of repetitive behaviors |  |  |  |

**Daily Activities**

| Sleeping less |  |  |  |
| Sleeping more |  |  |  |
| Eating less, not interested in food |  |  |  |
| Eating more than usual |  |  |  |
| Laying around, acting lethargic |  |  |  |

**Facial**

| Grinding teeth, clenching jaw down |  |  |  |
| Chewing, moving tongue around and/or out, smacking lips |  |  |  |
| Furrowing brow |  |  |  |
| Squinching, squinting eyes |  |  |  |
| Widening eyes |  |  |  |
| Frowning, turning down mouth |  |  |  |
| Pouting, quivering, puckering lips |  |  |  |
| Tensing face |  |  |  |
| Facial restlessness, tics |  |  |  |
| Grimacing |  |  |  |
| Lip biting |  |  |  |
| Continuous eye blinking |  |  |  |
| Rolling eyes |  |  |  |
| Flaring nostrils |  |  |  |
| Raising eyebrows |  |  |  |
| Lip curling |  |  |  |
Please feel free to offer comments, or recommend additional behavioral indicators that might not have been listed above.

Again, thank you.
Dear Participant,

I am a doctoral candidate in Nursing, at the University of Florida, and am inviting you to participate because of your experience in caring for children with autism spectrum disorders. Thank you for taking the time to provide your expert insight regarding how children with autism spectrum disorder (ASD) behaviorally express physical pain.

My research seeks to better understand how children with ASD express their physical pain, and aims to ultimately equip providers and caregivers with a tool that can be used to assess pain in this population, specifically in those children unable to verbally communicate their discomfort.

Because of the lack of information regarding how children with ASD express pain, and because of the confusion that surrounds this issue (given various reports of these children having a high tolerance for pain), it was determined that the development of a new pain assessment tool for this population is warranted; specifically, for those children with ASD who have severe communication and language impairments. Existing pain assessment tools have not been utilized and/or validated in this population, and there are limitations to using traditional pain assessment methods in children with ASD. Given the unique nature of this disorder, and the heterogeneity found in children on the spectrum, this population requires a novel approach to pain assessment.

The development of this tool is in the early stages. Recent work aimed to generate a long list of items that might indicate pain in children with ASD who cannot verbally communicate well. The current item pool you will review presents this list of behaviors. This list has been thoughtfully derived from several sources: the pain and autism literature, one-to-one discussion with several autism experts, semi-structured caregiver interviews, and caregiver surveys that asked about the pain experience in their children with autism. The next step in examining these theoretical items is to have a panel of experts review the potential indicators, and rate whether each one is relevant to this population. This will ensure that the revised set of items (which will undergo further review via a large test sample in the next step of instrument development) achieves adequate content validity. Your thought regarding whether each item should remain in the item pool at this stage in instrument development is crucial to ensuring that all potential indicators have been considered, and included.

Given the heterogeneous nature of this disorder, it is acknowledged that this (creating a universal pain assessment tool for this population) might not be possible, however it is believed that in attempting to gather information regarding how children with autism express pain, we can illuminate the fact that while some children might seemingly show an insensitivity to pain, many children on the spectrum do in fact feel and process pain, and therefore cannot be ignored or dismissed regarding appropriate pain management.

I sincerely appreciate your time, and am available should you have additional comments, insights, or questions.

Kindest Regards,

Melissa Dodd Inglese, Doctoral Candidate, ARNP
University of Florida College of Nursing
LIST OF REFERENCES


BIOGRAPHICAL SKETCH

Melissa Kay Dodd Inglese was born on May 27, 1979 in Yokosuka, Japan. The daughter of a Navy dentist, Melissa’s travels and early exposure to medical practices and health concerns all over the world prompted her to pursue a career in nursing. She graduated from the University of Florida in 2001, with a Bachelor of Science degree in nursing. Her developing passion for children led her to pursue advanced degrees in nursing through the accelerated BSN-PhD program offered by the University of Florida. In 2002, Melissa received her Master of Science degree in nursing and began her clinical work as a pediatric nurse practitioner, while simultaneously continuing to pursue her doctoral degree.

Melissa’s interests involve the alleviation of suffering in vulnerable populations of children. Specifically, her clinical work focuses on children with complex chronic conditions—conditions requiring children and families to navigate America’s vastly complex healthcare system. Her attention to proficient pain assessment and management in various pediatric populations led her to question how children with autism spectrum disorder (ASD)—a population receiving national and international attention—experience and express physical discomfort. Thus the focus of her doctoral work was directed towards this unique group of individuals. Melissa now continues her clinical and academic work focused on serving vulnerable populations of children.