THE EXPRESSION OF ACUTE PAIN REACTIONS IN CHILDREN WITH AUTISM: A COMPARATIVE ANALYSIS

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Abstract

The present study examined acute pain expressions of children with autism. It has been widely reported that reduced pain sensitivity is a common feature of children with autism. However, the evidence supporting this conclusion is based on anecdotal observations and studies using questionable measures of pain. Assessment of pain in children with autism is difficult due to self-report and nonverbal expression communication impairments. Caregivers often must provide proxy reports. The current study used objective behavioral measures of pain (facial activity and distress responses). Twenty-one children with autism were videotaped while receiving a venepuncture, with parental assessments of pain collected before and after the procedure. Twenty-two children without autism served as an age and gender matched control group. The results showed that children with autism display a significant behavioral response to the venepuncture procedure, with the response comparable to that observed in the control children. The concordance between parental reports of pain and observed pain responses of the child was consistently greater for the control group over the autism group. The results put into question the validity of parental global report as an assessment tool for pain in children with autism. The findings are explained using the sociocommunicative model of pain which views pain as not solely an internal experience, but as an interpersonal phenomenon and takes into account the encoding and decoding processes involved in the communication of pain.
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Introduction

Infantile autism is a pervasive developmental disorder with a prevalence of about five cases per 10,000 (American Psychiatric Association, 2000). It is characterized by an inability to form normal attachments and social relationships and is usually associated with impaired or absent verbal skills. Autism is also associated with cognitive impairment (75% of children with autism function at an intellectually disabled level) (Peterson, 1986). Despite the relatively low incidence of autism, it has received a considerable amount of media and scientific attention. Since the disorder was first described by Kanner in 1944 (Abelson, 1983), autism has been the subject of countless books and journal articles.

Despite the vast body of research on autism and the popular media fascination with the disorder, relatively little is known about the experience of pain in children with autism. The scientific and professional literature often reports pain insensitivity or indifference, but, to date, no empirical research has reported or documented the pain experience in these children. In fact, most of the reports of altered pain sensations in children with autism come from anecdotal observations and clinical impressions (e.g., Bettelheim, 1967; Wing, 1996).

Strangely, these conclusions have not been questioned or disputed. Rather, the view that children with autism experience pain insensitivity or increased pain thresholds is accepted as fact. The focus of most accounts of pain in children with autism has been aimed at explaining the mechanism behind the reduced pain sensitivity. Yet, in many ways, this puts the cart before the horse. Researchers are examining why children with autism have increased thresholds for pain before it has been shown that this is actually the case.

The present study empirically examined pain experience in children with autism using
both behavioral measures and parental reports of their child's pain during a venepuncture.
This was contrasted with the pain response in a group of children without autism experiencing
a similar medical procedure.

Literature Review

Pain Insensitivity in Autism and Cognitively Impaired Populations

The observation that children with autism have reduced pain sensitivity has been
frequently noted in the literature. Bettelheim (1967) described an account of a girl with
autism who showed no pain reactions despite having a high temperature and high white-blood
cell count. Appendicitis was suspected, but she showed no response to palpitation when she
was examined. She reportedly showed no pain reactions and did not protect the area.
However, she continued to become ever more sick until she fell into a coma. Emergency
exploratory surgery revealed a ruptured appendix, which was at least two days old.
Bettelheim (1967) concluded, "... it is difficult to imagine that anyone could stand the terrible
pain some of these children [children with autism] must have felt if they were experiencing
pain as normal people do" (p.58). But, there were no attempts to provide comparable data on
children without autism.

In recent years, several authors have described similar reports of reactions to
seemingly painful events in children with autism. Children with autism have been described as
having reduced pain sensitivity (Baranek & Berkson, 1994; Gillberg, 1995; Gillberg, Terenius
& Lonnerholm, 1985; Peeters & Gillberg, 1999) and not feeling pain as intensely as others
(Gillberg, 1988). Wing (1996) describes children with autism as having an "indifference to
pain" (p. 52), describing cases of children experiencing broken bones, dental abscesses and
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appendicitis and yet reacting as if nothing were wrong. Even the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV) identifies a high threshold for pain as associated with autism (American Psychiatric Association, 2000). None of these authors provided empirical data to support their conclusions.

The idea that people with cognitive or neurological impairment have reduced sensitivity for pain has been reported in the literature for almost half a century. Couston (1954) described seven cases of people with "mental deficits" who seemed indifferent to pain; they showed no observable signs of pain, but no systematic measurement system was employed. A literature review by Thrush (1973) reported that 28% of reported cases of congenital insensitivity to pain had associated mental retardation (although intelligence was not reported in all of the cases). Based on verbal, avoidance and psychophysiological measures, pain insensitivity in schizophrenia has been widely reported (Dworkin, 1994). Jancar and Speller (1994) conducted a retrospective study of hospital records over a fifty-year period examining the incidence of fatal intestinal obstruction in mentally handicapped people. Using data from 32 patients, they reported that fatal intestinal obstruction is more common in mentally handicapped people. Despite acknowledging the potential role of communicative deficits in the fatalities, they concluded, "Clinically, it is apparent that pain thresholds vary more widely in the mentally handicapped than in the general population" (Jancar & Speller, 1994, p. 420). Regrettably, no empirical basis for this broad generalization is provided.

Some of the most frequently cited work on the topic of reduced pain sensitivity in people with developmental disabilities has been done by Biersdorff (1991, 1994). She reported that the psychological literature on pain contains reports that pain insensitivity is a
common phenomenon in developmental disabilities and that the proportion of case studies of pain insensitive people, who also have developmental disabilities, is higher than one would expect. This is based on more than 80 case studies in the literature since the 1930's (Biersdorff, 1994). Despite acknowledging the role of cognitive limitations in terms of memory and conceptualization skills, Biersdorff (1991) argues, “One would expect at least some of the common responses to pain to be in the behavior repertoires of individuals with developmental disabilities regardless of articulatory constraints” (p. 360). However, Biersdorff’s conclusions were based on the findings of case studies which are highly selected, atypical reports dramatic enough to capture clinician, author and editor attention. To generalize from case studies to the entire population of individuals with developmental delays is inappropriate.

In 1994, Biersdorff conducted a study examining pain insensitivity and indifference in people with developmental disabilities. She used third party reporting of injury or illness incidents to estimate the incidence of pain insensitivity in 123 people with developmental disabilities. She found that 25% of the sample was reported to have a significantly elevated pain threshold. As well, those with more severe mental retardation were more likely to display insensitivity (Biersdorff, 1994). However, proxy reports of pain are notoriously unreliable and often under-estimate the child's self-report of pain when that is available (Chambers, Reid, Craig, McGrath, & Finley, 1998). As well, no control nor baseline data on people without developmental disabilities were provided.

More recently, work has been done to examine the pain responses of children with neurological impairment and developmental delays using objective, behavioral descriptions
rather than observer judgements. Oberlander, Gilbert, Chambers, O'Donnell and Craig (1999) found dampened behavioral (facial reactions) and physiologic (heart rate) reactions to a vaccination injection in eight profoundly disabled adolescents with significant neurological impairment. Gilbert-MacLeod, Craig, Rocha and Mathias (2000) examined everyday pain in developmentally delayed children. They found that the developmentally delayed children only exhibited moderate responses to everyday bumps and injuries and did not seek attention to the same degree as children without delays. However, unlike Biersdorff, Gilbert-MacLeod et al. suggested that the altered pain response could be due to a sociocommunicative deficit rather than a biophysical difference between the two populations (Gilbert-MacLeod et al., 2000).

**Pain Insensitivity or Pain Indifference?**

A serious concern when reviewing the literature on pain in people with developmental delays or cognitive impairment is the seemingly interchangeable use of the terms pain indifference and pain insensitivity. Congenital insensitivity to pain is a very rare phenomenon in the general population and is different from congenital indifference (Yanagida, 1978). Insensitivity and indifference differ in terms of the source of the problem in the nervous system (Biersdorff, 1994). In general, pain insensitivity refers to a decreased sensory pain experience, whereas pain indifference is a decreased emotional response to pain (Oberlander, O'Donnell, & Montgomery, 1999; Biersdorff, 1991). Pain indifference inhibits the pain experience from all sources, while pain insensitivity usually leaves the pain experience associated with internal organs relatively intact (Biersdorff, 1991). With pain indifference, the person has intact sensory pathways and peripheral nerves, but he or she fails to appreciate the painful nature of nociceptive input to the central nervous system. As a result, the person does not react with
flight behavior or defensiveness (Manfredi, Bini, Cruccu, Accornero, Berardelli et al., 1981):

 Pain insensitivity may result from abnormalities of the peripheral nerves or central sensory pathways. The absence of pain sensitivity has been speculated to be due to several factors: lack of receptors for nociceptive stimuli; impaired transmission in specific pain pathways; hyperactivity of pain controlling systems; and/or a defect of central integration of the category of pain (Manfredi et al., 1981).

The Opioid Hypothesis

Having acknowledged, perhaps inappropriately, that the pain experience in children with autism is different from other children, researchers set out to try to explain the difference. One of the main hypotheses involves the notion that children with autism experience pain insensitivity rather than pain indifference. Yanagida (1978) suggested that overproduction of brain endorphins in people with neurological impairment could be a potential cause of the apparent pain insensitivity. The opioid hypothesis has been tied to autism in particular.

Essentially, the opioid hypothesis suggests that hyperfunctioning of the endogenous opioid system could explain many of the symptoms associated with autism, including pain insensitivity (Gillberg, 1995). The search for reasons why children with autism ostensibly have this hyperfunctioning endogenous opioid system has triggered a series of hypotheses. The endogenous opioid levels may be increased for genetic reasons. They could result through primary overproduction, deficient degradation, abnormal messenger mechanisms within the nerve cells, or feedback deregulation mechanisms (Gillberg, 1995). Another hypothesis is that the repetitive behaviors commonly observed in children with autism (including self-injurious behaviors) may elevate the opioids. The stereotypic motor behaviors
involved in these repetitive activities could increase brain opioid levels, indirectly producing euphoria, some autistic symptoms and pain insensitivity (Gillberg, 1995).

Gillberg et al. (1985) provided early evidence for the opioid hypothesis in a study examining pain sensitivity and opioid levels in children with autism, children suffering from psychoses and a control group of children. Gillberg et al. measured pain sensitivity by asking the parents questions about how their children related to painful events in the past. The scores ranged from 1 (He or She is fussy) to 5 (Does not seem to react at all). Any children whose parents rated a 4 or a 5 were identified as having decreased sensitivity to pain. They measured the opioid levels by assaying endorphin levels in the cerebral spinal fluid (CSF) of the children. Gillberg et al. found a positive association between fraction II endorphin levels and decreased sensitivity to pain. They also reported that the fraction II endorphin levels were found to be higher in the autism and psychoses groups compared to the control group. A follow up study by Gillberg (1988) found many of the same results suggesting that reduced pain sensitivity (as rated by the parents) was associated with high fraction II endorphin levels and that children with autism have high levels of endorphin fraction II in their CSF.

However, a more recent study by Nagamitsu, Matsuishi, Kisa, Komori, Miyazaki et al. (1997) found different results. They found that CSF levels of beta-endorphin did not differ significantly between a group of children with autism and a strictly age-matched control group. As well, Nagamitsu et al. did not find any significant correlations between CSF beta-endorphin levels and clinical symptoms of autism (including self-injurious behavior and pain insensitivity). Nagamitsu et al. explained the differences between their findings and previous studies (such as Gillberg) by pointing out that the previous studies lacked an age-matched
control group. Gillberg et al. (1985) and Gillberg (1988) did not age match the comparison groups used. In fact, the control group used by Gillberg in each study consisted of only eight children (ages 6 months to 6 years old) with whom he compared 20 children with autism (ages 2 to 13 years old).

Despite a promising premise, the opioid hypothesis for autism has not yet been supported with empirical data. Even Gillberg (1995) admitted that the empirical study of the opioid hypothesis has produced few data of clinical relevance. The Gillberg (1985, 1988) studies seemed to provide support for the hypothesis, but suffered from methodological problems. The first problem was the lack of the strictly age-matched control group discussed earlier. As well, all of the studies cited above suffered from measurement problems; in particular, the measurement of pain sensitivity. The studies used retrospective parent report of painful events and their child's reaction to those events. A more direct, objective measure of pain reactivity would be desirable.

Sociocommunicative Deficits as a Possible Mechanism for Apparent Insensitivity

The evidence for reduced pain reactions in populations of cognitively or neurologically impaired people appears to be associated with the level of impairment. In general, as the level of the impairment increases, the pain response seemingly decreases (regardless of whether the measures are behavioral or physiological) (Oberlander, Gilbert et al., 1999). In a study of pain in elderly patients suffering from dementia, LaChapelle, Hadjistavropoulos and Craig (1999) found a similar association between the degree of cognitive impairment and the amount of pain reported verbally by the patients, but not in objectively coded nonverbal behavior. In children with autism, lower IQ levels are associated with greater self-injurious behavior (SIB)
(Bartak & Rutter, 1976). The greater level of SIB could suggest that the children with autism do not experience or react to pain in the same manner as others.

The association between cognitive/neurological impairment and apparent reduced pain sensitivity may suggest a possible mechanism for why these populations are thought to experience reduced pain sensitivity. In people with neurological impairment, cognitive, communicative and/or motor disabilities may become serious barriers to obtaining pain relief. "Children who consistently fail to display common pain behaviors when injured or ill may be thought to experience no or reduced pain" (Oberlander, O'Donnell et al., 1999, p. 235). The proposition that there is pain insensitivity in these populations could have a basis in several reasons: a) failure of others to appreciate communication deficits, b) caregivers' misperceptions of the children, or c) lack of systematic study of the problem (Oberlander, Gilbert et al., 1999). Despite the widely reported belief that children with cognitive impairment experience reduced pain sensitivity, no detailed studies examining behavioral reactions have been reported. "There is no documented or systematic reason to believe that people with communicative or cognitive limitations are spared the suffering of pain" (Oberlander, O'Donnell et al., 1999, p. 236).

An alternative explanation for the apparent pain insensitivity in children with autism is a sociocommunicative one. The reductions in communication skills and social relatedness are what most consistently distinguish children with autism from children without autism (DeMyer, Hingtgen, & Jackson, 1981). In fact, Kanner, who was the first to document the disorder, regarded social dysfunction and the unusual responses to the environment as the two essential features of autism (Grossman, Carter, & Volkman, 1997).
The cognitive impairments found in children with autism would traditionally be considered mediated by the left hemisphere. These include verbal, sequential processing and analytic skills which are all almost uniformly poor or absent in children with autism (Grossman et al., 1997; Prior, 1979). Skills mediated by the right hemisphere (e.g., visuospatial skills) are much less handicapped in children with autism (Prior, 1979). It is common for a delay, or total lack of development of spoken language to characterize this population (Abelson, 1983; American Psychiatric Association, 2000). If speech does develop, it tends to be atonal, lacks inflection and fails to convey emotions (Peterson, 1986). Their language is characterized by concreteness, repetitiveness and a mechanical/noncommunicative approach (Prior, 1979). Therefore, if language does develop in these children, it tends to be robotic and unemotional.

Autism is also characterized by an impairment in nonverbal and social behaviors. Abelson (1983) reported that autism seems to be characterized by a failure to develop social relationships rather than a withdrawal from relationships. So the children do not withdraw from relationships that have been formed, rather, they do not form relationships in the beginning. Children with autism tend to have poor or deviant eye contact, an absent or delayed social smile and impairment in the use of other facial expressions (Abelson, 1983; American Psychiatric Association, 2000; Peterson, 1986). This could partially explain the failure of these children to form relationships with parents and other caregivers. They tend to have a pervasive lack of responsiveness to other people. They prefer to be alone and may seem to ignore humans while attending to nonhuman objects in the environment (Brooker & Mareth, 1982; Peterson, 1986).

This lack of social functioning and impaired language use could explain the apparent
pain insensitivity in children with autism. The substantial impairment in all language functions (verbal and nonverbal) could influence how parents and other caregivers interpret the responses to a seemingly painful stimulus in children with autism. For example, if the child does not cry or seek comfort from a parent after an injury, it could be inferred that the child is not experiencing pain. However, these children do not form strong bonds with people and may seek comfort from an inanimate object in the environment. Even if the children did approach their parents, they would not be able to communicate their pain experience.

Children with autism tend to have difficulty transferring information from one sense modality to another and then integrating that information. They also have difficulties in the use of body gestures and understanding the receptive language of others (Abelson, 1983). Therefore, it is not surprising that these children do not seem to express pain or discomfort to others.

Does the Definition of Pain Apply to Cognitively Impaired Populations?

One of the contributing factors to the prevailing view that children with cognitive impairment have decreased sensitivity for pain could be the definition of pain itself. Pain was defined by the IASP Committee for Taxonomy as, “An unpleasant sensory and emotional experience associated with actual or potential tissue damage, or described in terms of such damage. . . . pain is always subjective. Each individual learns the application of the word through experiences related to injury in early life” (Merskey, 1991, p. 153). Unfortunately, this definition is inadequate for cognitively impaired children, nonverbal elderly, preverbal infants and organisms incapable of verbal report (Anand & Craig, 1996; Oberlander, O'Donnell et al., 1999).

Such a definition could result in the discounting, neglect, and substandard treatment of
pain in these populations (Anand and Craig, 1996; Oberlander, Gilbert et al., 1999). Parents and health care professionals may discount or deny signals of acute distress in children who cannot clearly express their pain (Walco, Cassidy, & Schechter, 1994). This definition of pain relies on the assumption that the experience of pain and the reporting of pain are congruent, but this may not be the case in people who have verbal limitations (Anand & Craig, 1996). The requirement of verbal report may result in the failure to recognize and adequately treat pain in children with cognitive impairment.

Oberlander, O’Donnell et al. (1999) point out another problem with the IASP definition of pain. The definition depends on the previous experience of pain. However, Oberlander, O’Donnell et al. (1999) argue that the perception of pain is common in all organisms with a nervous system and therefore, the perception of pain should not depend on past painful experiences. The first experience of tissue damage should be just as painful as subsequent experiences. What will change is the interpretation and meaning of the painful sensations over time (Oberlander, O’Donnell et al., 1999). Cognitively impaired children who may have difficulty relating tissue damage to past experiences should not be thought of as not experiencing pain.

Pain Assessment in the Cognitively Impaired

Several studies have examined how parents of children with cognitive or neurological impairments determine if their child is in pain. A recent study by Fanurik, Koh, Schmitz, Harrison and Conrad (1999) examined this topic using the parents of a heterogenous group of cognitively impaired children. Fanurik, Koh, Schmitz, Harrison and Conrad (1999) asked the parents questions like, “How do you know when your child is in pain?” and “Do you think
your child feels or experiences pain in the same way as a child who is not delayed or impaired?" (p. 230). Parents reported that the majority (66%) of mild to moderately impaired children were directly able to express their pain through self-report. However, 90% of the parents of severely/profoundly impaired children reported that they were only able to detect the presence of pain through the child's behavior (vocalizations, eating/sleeping, activity levels). These parents could not use their child's verbal report of pain. Regardless of the level of cognitive impairment, the majority of parents reported that they believed that their children experience pain differently than children without cognitive impairment. They reported that they perceived decreased pain sensitivity and increased pain tolerance in their children compared to other children. However, the study did have problems which Fanurik, Koh, Schmitz, Harrison and Conrad pointed out. The cognitive level of the child was based solely on parental report and that may not have been an accurate measure of the actual level of functioning of the child. Also, the questions may have been leading (especially the question comparing the child's pain experience to that of non-impaired children). However, a bigger problem had to do with what the parent was reporting as the pain experience of the child. Fanurik, Koh, Schmitz, Harrison and Conrad reported that they were uncertain as to whether the parents were referring to pain tolerance, pain sensitivity or both. As well, there could be a diminished capacity to interpret and understand the meaning of the experience, but an unaltered capacity for the severity and emotional distress of pain. Detailed examination of the nature of pain reactions is needed.

Another study examined the behaviors that caregivers used to assess pain in patients with cerebral palsy (McGrath, Rosmus, Canfield, Campbell, & Hennigar, 1998). McGrath et
al. asked caregivers to recall instances of acute pain and to describe the person's reactions to them. Since many people with cerebral palsy have behavioral limitations and idiosyncracies that may mask pain expression, the typical pain indicators may be inconsistent and difficult to interpret in patients with cerebral palsy (McGrath et al., 1998). What they found was that they could not use a single set of items or behaviors to discriminate pain in this population. However, although the exact topology of the reactions differed across individuals, there were commonalities among the behaviors caregivers reported as indicative of pain. For example, 80% of caregivers reported that they used aspects of crying and moaning to alert them to possible pain (McGrath et al., 1998).

A study by Fanurik, Koh, Schmitz, Harrison, Roberson and Killebrew (1999) looked at the attitudes of health care providers toward pain assessment and treatment in people with cognitive impairment. They found that physicians and nurses' views toward analgesia and pain management were similar in children with or without cognitive impairment. Fanurik, Koh, Schmitz, Harrison, Roberson et al. did point out that it was unclear whether these views toward pain management were consistent with clinical practice. They cite a previously mentioned study (Fanurik, Koh, Schmitz, Harrison, & Conrad, 1999) which reported that 26% of parents of children with cognitive impairment felt that health care providers underestimated and/or under treated their child's pain. However, the presence of cognitive impairment did influence the health care providers' decisions regarding pain assessment. Self-report was considered less appropriate for assessing pain in this population (Fanurik, Koh, Schmitz, Harrison, Roberson et al., 1999).

The issue of pain assessment in this population is a complicated one. In general,
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report is considered the "gold standard" for assessing pain (Fanurik, Koh, Harrison, Conrad & Tomerlin, 1998; McGrath et al., 1998), although this claim overstates its merits. As has already been pointed out, self-report is usually unavailable or unreliable when dealing with children with profound cognitive or neurological impairment. When available, the overall validity of self-report in children with cognitive impairment is questionable (Fanurik, Schmitz, Harrison, Roberson et al., 1999). “In general, pain assessment in children can be subjective and is shaped by cognitive and language capacities inherent to children” (Oberlander, O’Donnell et al., 1999, p. 238). When the child is nonverbal and cognitively impaired, the child has a limited repertoire of distress signals and these are often ambiguous with respect to the source. The child could appear to be experiencing pain, fear, sadness or even contentment (Oberlander, O’Donnell et al., 1999). Many children with cognitive impairment have idiosyncratic behaviors like vocal abnormalities or facial peculiarities. This could result in overestimates of pain by caregivers unfamiliar with the child (Fanurik, Koh, Schmitz, Harrison, & Conrad, 1999), although this rarely seems to happen. As a result, in this population, pain should be assessed by understanding the child’s behavior in the context of past experiences. The “pain” behavior should be compared with known baseline behavior for the child in order to assess the level of pain (Oberlander, O’Donnell et al., 1999). Since there are currently no good pain measures specific for this population, multiple sources of information should be used to assess pain in these children (Oberlander, O’Donnell et al., 1999). These may include developmentally appropriate pain rating scales, behavioral observations and physiological measures (Fanurik, Koh, Schmitz, Harrison, & Conrad, 1999). Of course, in the absence of validity data, additional measures could only be adding noise to
the data available. For example, physiological measures are often seen as good, objective measure of pain. However, in general, physiological indices like heart rate and blood pressure are non-specific and may reflect illness or general stress. The physiological measures most often employed are not specific to pain and may reflect several possible causes.

Some verbal report scales have been developed for use in this population, although with limitations. Fanurik et al. (1998) developed a 0-5 numerical scale for assessing pain in children, aged 8-17, with cognitive impairment. However, the scale was only useful with low levels of impairment, according to their standards. Fanurik et al. reported that 50% of borderline and 35% of moderately impaired children could correctly use the scale. As well, nurses overestimated the abilities of the children to use the scale. A study by Bromley, Emerson and Caine (1998) had people with intellectual disabilities respond to a series of photographs of painful experiences. They then had the participants rate pain intensity and location using a body map. Bromley et al. found that people with intellectual disabilities could reliably indicate pain location on the body map and rate pain intensity using an analogue color scale. However, it is unclear whether or not the participants would be able to rate their own pain; it could be that it is different to rate pain when the rater is not the one experiencing it.

The reliability issues concerning verbal report demand a more objective assessment measure for pain in the cognitively impaired. LaChapelle et al. (1999) conducted a study examining the use of facial expressions to assess pain in cognitively impaired adults receiving a vaccination. In their study, they found that 35% of the participants could not provide verbal report and even those that could provided reports of questionable reliability. In participants who could provide verbal report, LaChapelle et al. found no significant differences in the
verbal report pain ratings between baseline and injection phases. However, they did find that the intensity of facial activity of the participants showed increases from the baseline to injection phases. This was also the same pattern for observer judgements for the baseline and injection phases. LaChapelle et al. found no significant correlation between the frequency and intensity of facial activity and level of cognitive functioning. They concluded that the large proportion of individuals unable to provide verbal report, as well as the questions about the reliability of verbal report when available, supported the need for a more objective nonverbal measure of pain. LaChapelle et al. also concluded that nonverbal facial expression is an important indicator of pain in this population. Using facial expression, they found no evidence for insensitivity to pain in adults with cognitive impairment and suggested that the deficit may be in their ability to communicate their pain experience to others.

However, a study by Oberlander, Gilbert et al. (1999) provided somewhat different results. Oberlander et al. examined the facial reactions of eight adolescents with significant neurological impairment to mock and actual vaccinations. They found small or no differences in the pain response across the mock and real vaccine conditions. The facial reaction to the actual injection was dampened from what was expected. However, Oberlander et al. warned that the results should not be used to conclude that adolescents with neurological or cognitive impairment do not experience pain. Rather, they suggest that the neurological impairment may blunt the pain response. Oberlander et al. also point out that the subjects were autonomically aroused before the injection took place. The holding and preparation of the patient for the injection could have evoked arousal similar to the actual injection. Since the patients were already autonomically aroused, they perhaps could not mount a further arousal
Oberlander, Gilbert et al. (1999) focused their conclusions around the idea that an altered pain display in this population does not necessarily suggest pain insensitivity. The perception of pain (afferent component) must be experienced before behavioral (efferent) responses can occur (Oberlander, O’Donnell et al., 1999). However, the fact is, very little is known about the integrity of the nociceptive system in children with cognitive impairment (Fanurik, Koh, Schmitz, Harrison, & Conrad, 1999). Therefore, concluding that any reduction in pain display is evidence of pain insensitivity in cognitively impaired children or adults is premature. Neurological or cognitive impairment may alter the ability to comprehend and communicate pain, but there is no evidence that this reflects true pain insensitivity (Oberlander, O’Donnell et al., 1999). The seemingly altered pain expression often seen in this population could also be due to medical personnel and caregivers being unaware or unable to interpret the expression accurately (LaChapelle et al., 1999). The differences between the perception and expression of pain depend on the neurological condition and the environmental context of the patient (Oberlander, O’Donnell et al., 1999). This is especially apparent in children with autism. Any apparent decrease in pain reactivity observed in children with autism does not necessarily result from insensitivity, but rather, may represent a different mode of pain expression. This different mode of pain expression would be related to difficulties with verbal communication, nonverbal behavior and certain cognitive disorders (problems representing sensations and emotions) (Tordjman, Antoine, Cohen, Gauvain-Piquard, Carlier et al., 1999).
Behavioral Pain Assessment

The first purpose of this study was to characterize the behavioral responses of children with autism experiencing a painful medical procedure (venepuncture). As described earlier, the research literature has suggested that children with autism display reduced pain sensitivity. However, much of the evidence supporting this is based on anecdotal observations and studies using poor measures of pain. The pain experience in children with autism was examined using objective observational measures of pain and distress. The reaction to the invasive venepuncture procedure was contrasted with baseline and pre-procedure measures of pain and distress to determine the children's reactions to the noxious event.

The first of these measures was the Child Facial Coding System (CFCS; Chambers, Cassidy, McGrath, Gilbert, & Craig, 1996). CFCS is a facial coding system designed to assess the pain experience in preschool aged children. The system codes for 13 facial actions adapted from the Facial Action Coding System (FACS; Ekman & Friesen, 1978) and the Neonatal Facial Coding System (NFCS; Grunau & Craig, 1987): brow lower, squint, eye squeeze, blink, flared nostril, nose wrinkler, nasolabial furrow, cheek raiser, open lips, upper lip raise, lip corner puller, vertical mouth stretch and horizontal mouth stretch (Chambers et al., 1996). CFCS has been used to assess pain responses to both acute (Oberlander, Gilbert et al., 1999) and persistent pain (Gilbert, Lilley, Craig, McGrath, Court, et al., 1999).

The second measure used was the Observational Scale of Behavioral Distress - Revised (OSBD; Jay & Elliott, 1984). OSBD is based on the Procedural Behavior Rating Scale (PBRS) developed by Katz, Kellerman and Siegel (1980). The PBRS was designed to assess behavioral distress in children undergoing painful bone marrow aspirations. Jay and
Elliott (1984) made two major revisions to the PBRS to derive the OSBD. First, the distress behaviors are recorded in continuous set intervals within each phase of the medical procedure, rather than one gross recording of occurrence over the entire phase. Secondly, each behavioral category is weighted according to the intensity of the behavior (Elliott, Jay, & Woody, 1987). OSBD consists of eight operationally defined behaviors indicative of anxiety and/or pain behavior in children: information seeking, cry, scream, restraint, verbal resistance, emotional support, verbal pain and flail (Jay & Elliott, 1986). It is a reliable and valid measure of children's distress and can be used to measure children's distress in stressful medical situations like injections and venepuncture (Jay & Elliott, 1984).

Comparison Between Children with Autism and Non-Cognitively Impaired Children

The second purpose of this study was to compare the behavioral reactions of children with and without autism undergoing venepuncture. One of the major methodological problems with other studies examining pain in children with autism and other cognitive impairments is that most of the studies lack a proper control group. This author has found no empirical studies comparing pain expression in children with autism and children without autism. Comparing the behavioral expressions of pain (measured by CFCS and OSBD) of children with and without autism would provide empirical evidence for or against the conclusion that children with autism experience pain differently than children without autism.

Parents' Reports of Pain in Children

The third purpose of this study was to examine parents' assessments of pain behavior in children with autism and children without autism. As discussed earlier, Fanurik, Koh, Schmitz, Harrison and Conrad (1999) explained that parents of children with cognitive
impairment reported that they felt their children experience pain differently than other children. If this were the case, one would expect that parents of children with autism would assess pain differently than parents of children without autism. Any behavioral sources of differences between the two groups of parents could be examined by comparing the concordance between parental report and their child’s expression of pain.

The parental report of pain was measured using the Faces Pain Scale (Appendix A) (FPS; Bieri, Reeve, Champion, Addicoat, & Ziegler, 1990). The FPS consists of seven faces showing gradual increases in pain expression from left to right (neutral to pain). The FPS is commonly cited in empirical literature and is widely used in clinical practice (Chambers, Giesbrecht, Craig, Bennett, & Huntsman, 1999). The scale was chosen because it has minimal cognitive demands and can be reliably and validly used in young children (Bieri et al., 1990). The FPS was chosen in an attempt to get self-report of pain in both the children with autism and the children without autism as well as parental reports of their children’s pain. In addition, as Oberlander, O’Donnell et al. (1999) explained, in children with cognitive impairment, pain can best be assessed by understanding the child’s behavior in the context of past experiences. The sensitivity to pain of the children was assessed by having the parents of children with autism and children without autism report on past pain reactions of their children. As reported earlier, Biersdorff (1994) found that third party reporting of pain in people with cognitive impairment suggested that they had higher pain thresholds. It is possible that retrospective third party reporting of past pain and the observed pain behavior during the painful event could be discordant. The parent reports of their child’s pain behavior were measured using the Dalhousie Pain Checklist (Appendix B) (DPC; McGrath et al., 1998).
which is a 30-item checklist of behaviors that caregivers use to determine pain in people with cognitive impairment. Although the DPC is a relatively new scale, it has been shown to be a valid and reliable measure of pain in this population (Breau, McGrath, Camfield, Rosmus, & Finley, 2000). In addition, the parents were asked to provide a report of their child's pain temperament by responding to the following statement: "My child is very sensitive to pain of bumps or cuts or other common hurts" (Appendix B) (Grunau, Whitfield and Petrie, 1994). The parent responded to this question on a scale of 1 = not typical/characteristic to 5 = very typical/characteristic. This question has been used by Grunau et al. to assess pain temperament in toddlers.

Hypotheses

1) The facial expressions and distress responses of the children with autism would not be significantly different from those of the children without autism.

2) The children with autism would show significantly more vigorous facial expressions and distress responses during the injection phase of the venepuncture procedure than during the noninvasive, baseline preparation.

3) The parents of the children with autism would report that their children are less responsive to pain using retrospective reports compared to the retrospective reports of parents of children without autism.

4) The parents of children with autism would report that their children are less sensitive to pain compared to the reports of parents of children without autism.

5) The concordance between parental report and observed expressions of pain would be significantly different between parents of children with autism and parents of children without
Methods

Participants

Twenty-one children with autism were recruited from a study already underway at Sunny Hill Health Centre for Children. The sample of children with autism consisted of 18 boys and three girls with a mean age of 5.42 years, $SD = 1.13$ years (range 3.17 – 7 years). The children and their parents were taking part in a study examining the effects of a hormone on autism. The hormone was administered via an intravenous injection. All of the children met DSM-IV diagnoses of autism, meeting an average of 8.95 criteria, $SD = 1.76$ (range 6 – 12 criteria) out of 12 possible diagnostic criteria. The degree of autism was assessed using the Childhood Autism Rating Scale (CARS; Schopler, Reichler, & Renner, 1998). The mean CARS score for the autism group was 39.10, $SD = 4.98$ (range 30.5 – 47) which put the group into the severely autistic range (CARS score greater than 37; Schopler et al., 1998). Breaking down the group, nine children fell into the mildly-moderately autistic range (CARS score 30 – 37) and 12 children fell into the severely autistic range (Schopler et al., 1998). The parents of the children with autism also completed the Autism Behavior Checklist (ABC; Krug, Arick, & Almond, 1980). The mean ABC total score was 76.3, $SD = 19.2$ (range 38 – 121). The ABC also breaks down into five symptom areas: sensory, relating, body and object use, language and social. The ABC scores for the autism group were: sensory = 12.8, $SD = 5.5$ (range 3 – 21); relating = 22.2, $SD = 6.6$ (range 8 – 33); body and object use = 14.1, $SD = 8.5$ (range 0 – 34); language = 14.0, $SD = 5.5$ (range 2 – 23); and social = 13.3, $SD = 4.8$ (range 5 – 21). All of the ABC scores for the autism group were similar to norms reported for
children with autism (Krug et al., 1980). All parents in the study were English speaking.

The control children were recruited through the British Columbia Children's Hospital outpatient blood collection laboratory. They were age and gender matched to the children with autism. The control group consisted of 22 children (18 boys, 4 girls) with a mean age of 5.16 years, SD = 1.33 years (range 3.10 – 7.85 years). Children with developmental delay or cognitive impairment were excluded from the study. The children were informally screened for cognitive impairment by having the parents complete a short questionnaire (Appendix C). The questionnaire asked about the child's relevant medical history (presence of central nervous system disorder, history of head injury or anoxia) and if teachers, daycare workers, or parents themselves had suspected potential delays (Franzen & Berg, 1989). Children experiencing any of these were excluded. Children currently on analgesic medication were also ineligible for participation. All parents and children were English speaking.

Procedure

The study was approved by the University of British Columbia Research Ethics Board and the Children's and Women's Health Centre of British Columbia Research Review Committee. For the children with autism, the families were sent a contact letter and informed consent form (Appendix D) by the researchers of the hormone study. In the informed consent form, it was made clear that if they chose not to participate in this study, they would still be able to participate in the hormone study. As well, the parents were also sent the DPC to complete beforehand. If parents were interested in participating in the study, they were asked to send back the signed informed consent form and bring the completed DPC with them on the day of the procedure. All demographic and psychological testing information was
collected afterwards from the files of the hormone study.

On the day of the injection, the parents were led to the procedure room by a nurse who introduced the parents to the researcher who would be videotaping the procedure. Videotaping was completed in the procedure room. The children were videotaped for a baseline period (approximately 3-5 minutes) before the procedure began. A randomly selected 10 second segment from this period served as the “baseline” phase for coding. The nurses and parents then bundled and restrained the child as the physician prepared the injection site. The bundling and restraint visibly produced a state of distress in the child, thereby providing a non-noxious, but aversive control contrast with the venepuncture itself. The 10 seconds immediately before the injection served as the “pre-needle” phase for coding. The 10 seconds immediately after the needle insertion was the “needle” phase. The procedure usually took approximately three minutes for the injection to be completed. However, some children required several tries to properly insert the needle. The procedure was videotaped in its entirety. The 10 seconds immediately after the needle was removed was the “post-needle” phase. After the child was unbundled and free to move, the researcher continued to videotape the child for approximately three minutes; from this period, a randomly selected 10 second segment was chosen as the “return to baseline” phase. The child and parents were then taken to another room where the parents completed the FPS. The FPS was completed by the mother in 17 of the cases and by the father in the remaining 4 cases. Attempts were made to have the children with autism complete the FPS, but the children were too distressed or impaired to provide a self-report.

For the control children, the parents were approached by a researcher upon arrival at
the blood collection lab. The parents were explained the study and given an information letter and informed consent form (Appendix E). If the parents agreed to participate, the researcher asked them the questions on the brief medical/cognitive history questionnaire (Appendix C). If the parent responded "yes" to any of the questions, they were asked to provide more details. Any children whose parents suspected or knew of developmental delay were excluded from the study; the parents were thanked for their time and no other information was collected. If the questionnaire was completed without any delay identified, the parents were then given the DPC to complete as they waited for their child to be called into the procedure room. When the children were called in, the researcher accompanied the child and parent into the procedure room and began videotaping of the child immediately. The videotaping was conducted in the same fashion as was done with the autism group. However, the procedure took much less time with the child only being videotaped for approximately 3-5 minutes, on average. After the procedure was completed, the parents were asked to rate the child's pain using the FPS. The FPS was completed by the mother in 16 cases and by the father in 6 cases. The children were also asked to rate their pain during the procedure, with only one child being too distressed to provide a self-report.

The videotapes were CFCS coded for the five segments (baseline, pre-needle, needle, post-needle, return to baseline) by trained coders who had achieved high reliability in training. The segments were coded using still motion, frame by frame examination. All of the 13 CFCS action units were coded for each second of the 10-second segments. This resulted in 10 scores for each CFCS action unit per coding segment. The reliability of the coders was checked by having one coder code all of the segments and another coder code 20% of those
segments randomly selected. A similar procedure was used for the OSBD coding.

OSBD coding was conducted slightly differently. The same 10-second segments that were coded with CFCS were coded with OSBD, but the entire 10-second segment was coded as a single unit, resulting in one score for each OSBD behavior per segment.

Data Analysis

Data Screening and Scoring

Prior to data analysis, dependent variables were examined for accuracy of data entry, missing values, and fit between their distributions and the assumptions of multivariate analyses.

CFCS data preparation. CFCS scores for each action unit per segment were obtained by calculating the average of the individual action unit scores for each segment. An overall facial action score for each segment was then generated by summing up the average scores for each of the action units in each segment.

OSBD data preparation. OSBD scores for each distress behavior per segment were obtained in a similar fashion; the average of the individual distress behavior scores for each segment was calculated. An overall behavioral distress score was calculated by summing up the average scores for each of the distress behaviors in each segment.

DPC scoring. The DPC score was calculated by summing up the scores on each of the thirty items on the checklist. Any missing data items were replaced by interpolation based upon the values which preceded and followed each data point (Breau et al., 2000).

Operational Definition of Pain Response. The only difference between the pre-needle and needle segments was the insertion of the needle during the needle segment. Therefore, the
pre-needle segment could serve as an anxiety/no pain control with which the needle segment could be compared. Any increase in facial activity or behavioral distress over and above the levels during the pre-needle segment could therefore be attributed to the needle insertion. As a result, facial pain response was operationalized as the difference between the overall CFCS scores for the needle and pre-needle segments. Using a similar rationale, behavioral distress pain response was operationalized as the difference between the overall OSBD scores for the needle and the pre-needle segments.

Results

Coding Reliability

Inter-rater scoring reliability for this sample was calculated using the formula recommended by Ekman and Friesen (1978) which assesses the proportion of agreement on actions recorded by two coders relative to the total number of actions coded as occurring by each coder. The inter-rater reliability for the CFCS coding of the autism group was 0.73 while the reliability for the control group CFCS coding was 0.78. The differences in reliability can be explained by differences in the position of the autism and control children during the venepuncture procedure. The children with autism were lying down which made coding more difficult compared to the control children who were sitting up in a chair. The different camera angles for the two groups likely resulted in the differences in inter-rater reliability. The inter-rater reliability is acceptable given the difficult coding conditions. The inter-rater reliability for the OSBD coding of the autism group was 0.84 while the reliability for the control group OSBD coding was 0.88.
CFCS Multivariate Analysis

A 2 (group: autism versus control) X 5 (coding segment: baseline versus pre-needle versus needle versus post-needle versus return to baseline) multivariate analysis of variance (MANOVA) with repeated measures was conducted to determine if there were differences between overall facial activity across any of the segments and between the autism and control groups. A multivariate main effect was found for the coding segments, $F(4, 164) = 24.64, p < 0.001$ (see Table 1 for descriptive statistics and Figure 1 for a graphical summary using the total facial action score). Student Newman Keuls post hoc tests showed that there was a significant difference in facial activity between the needle segment and all of the other segments, with the children showing the greatest facial activity during the needle segment. As well, there was a significant difference in facial activity between the pre-needle segment and all of the other segments with the children with autism evidencing a more vigorous reaction (see Table 3).

There was no significant multivariate main effect between the groups, $F(1, 41) = 2.75, p > 0.05$. However, a significant multivariate interaction between facial activity and group was observed, $F(4, 164) = 4.59, p < 0.01$. Univariate t-tests were conducted to detect simple main effects between the groups for each segment. The alpha level was set at $0.05/5 = 0.01$ to reduce the risk of Type 1 error. The only significant difference between facial activity for the autism group and control group occurred during the needle segment, $t(41) = 3.16, p < 0.01$, two-tailed (see Table 3).

OSBD Multivariate Analysis

A similar 2 (group: autism versus control) X 5 (coding segment: baseline versus pre-
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needle versus needle versus post-needle versus return to baseline) MANOVA with repeated measures was conducted to determine if there were differences between overall behavioral distress across any of the segments and between the autism and control groups. A multivariate main effect was found for the coding segment, $F(4, 164) = 33.20, p < 0.001$ (see Table 4 for descriptive statistics and Figure 2 for a graphical summary using the total OSBD score). Student Newman Keuls post hoc tests showed that there were significant differences in behavioral distress between the needle segment and baseline, post-needle, and return to baseline segments. There were significant differences in behavioral distress between the pre-needle segment and the baseline and return to baseline segments. Finally, there were significant differences in behavioral distress between post-needle and all of the other segments (see Table 5).

There was a significant multivariate main effect between the autism and control groups, $F(1, 41) = 23.68, p < 0.001$, with the children with autism displaying greater behavioral distress overall than the controls. In addition, a significant multivariate interaction was observed in behavioral distress between group and coding segment, $F(4, 164) = 9.65, p < 0.001$. Univariate t-tests were conducted to detect simple main effects between the groups for each segment. Again, the alpha level was set at $0.05/5 = 0.01$ to reduce the risk of Type 1 error. Significant differences were found between the autism group and control group (with the autism group displaying greater behavioral distress) during the pre-needle segment, $t(41) = 3.95, p < 0.001$, two-tailed; needle segment, $t(41) = 3.76, p < 0.01$, two-tailed; post-needle segment, $t(41) = 6.86, p < 0.001$, two-tailed; and return to baseline segment, $t(41) = 3.044, p < 0.01$, two-tailed (see Table 6).
Autism CFCS and Facial Activity Analysis

A five-level analysis of variance (ANOVA) with repeated measures was performed to examine if the children with autism showed a significant increase in facial activity in response to the venepuncture. The overall facial activity of children with autism differed across segments, $F(4, 80) = 25.75, p < 0.001$ (see Table 1 for descriptive statistics and Figure 1 for a graphical summary). Student Newman Keuls post hoc tests indicated a significant increase in facial activity between the pre-needle and needle segments, $p < 0.001$ (see Table 7).

To determine the pattern of facial activity associated with the venepuncture, a procedure similar to one described by LaChapelle et al. (1999) was followed. Facial actions were considered characteristic of pain if a) they occurred on more than 5% of the occasions in which they could have been observed in the needle segment and b) they occurred significantly more during the needle segment than during the pre-needle segment. Seven of the facial actions met these criteria: brow lower ($p < 0.01$), eye squeeze ($p < 0.05$), flared nostril ($p < 0.05$), nose wrinkler ($p < 0.01$), cheek raiser ($p < 0.01$), vertical mouth stretch ($p < 0.01$), and horizontal mouth stretch ($p < 0.05$).

Control CFCS and Facial Activity Analysis

A five level repeated measures ANOVA was performed to examine if the control children showed a significant increase in facial activity in response to the venepuncture. The facial activity of the control children did differ across segments, $F(4, 84) = 4.06, p < 0.01$ (see Table 1 for descriptive statistics and Figure 1 for a graphical summary). However, Student Newman Keuls post hoc tests failed to show a significant increase in facial activity between the pre-needle and needle segments (see Table 8). This finding is likely due to reduced power.
because of a small sample size. The difference between facial activity in the pre-needle and needle segments is not significant at alpha = 0.05, but is significant at alpha = 0.10.

An analysis (identical to the one done with the autism group) of the pattern of facial activity associated with the venepuncture was performed with the control group. In the control group, only three facial actions met the criteria outlined above: brow lower (p < 0.01), squint (p < 0.01), and flared nostril (p < 0.05).

**Autism OSBD Analysis**

A five level repeated measures ANOVA was performed to examine if the children with autism showed a significant increase in behavioral distress in response to the venepuncture. The behavioral distress of the children did differ across segments, $F(4, 80) = 36.32$, $p < 0.001$ (see Table 4 for descriptive statistics and Figure 2 for a graphical summary). The major difference was between the baseline and other segments. Student Newman Keuls post hoc tests failed to show a significant increase in behavioral distress between the pre-needle and needle segments (see Table 9).

**Control OSBD Analysis**

A five level repeated measures ANOVA was performed to examine if the control group of children displayed a significant increase in behavioral distress in response to the venepuncture. The assumption of sphericity was violated and as a result, a Greenhouse-Geisser correction for degrees of freedom was used. The behavioral distress of the children did differ across segments, $F(2.54, 53.29) = 6.552$, $p < 0.01$ (see Table 4 for descriptive statistics and Figure 2 for a graphical summary). Again, this was attributable to differences between the baseline and other segments. Similar to what was observed in the autism group,
Student Newman Keuls post hoc tests failed to show a significant increase in behavioral distress between the pre-needle and needle segments (see Table 10).

**Autism versus Control Facial and Behavioral Pain Response**

As discussed earlier, the operationalized definition of facial pain response was the facial activity during the needle segment minus the facial activity during the pre-needle segment. Any increase in facial activity was attributed to the noxious stimulus of the venepuncture. The mean facial pain response for the autism group was 3.53, SD = 3.96 and the mean facial pain response for the control group was 1.80, SD = 3.85. There was no statistically significant difference between the facial pain responses of the autism and control groups, \( t(41) = 1.46, p > 0.05 \).

The behavioral distress pain response was operationalized in a similar fashion: the difference between the overall OSBD scores for the needle and the pre-needle segments. The mean behavioral distress pain response for the autism group was 0.24, SD = 0.70 and the mean behavioral distress pain response for the control group was 0.36, SD = 1.29. The variances between the two groups were not equal and therefore a correction was used when calculating the degrees of freedom. There was no statistically significant difference between the behavioral distress pain responses of the autism and control groups, \( t(32.7) = -0.398, p < 0.05 \).

**Parental Assessment of Pain Reactivity and Temperament**

Parental assessment of pain reactivity in their children was measured using the total score from the DPC. The mean DPC score provided by the parents of children with autism was 60.33, SD = 13.50 while the mean DPC score provided by the parents of the control
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children was 58.41, SD = 14.19. The mean DPC scores between the autism group and the control group were not significantly different, t (41) = 0.46, p > 0.05.

Parent views of child pain temperament were assessed by having the parents complete the Grunau et al. (1994) pain temperament item. For the autism group, three of the 21 parents failed to respond to this item. As a result, the analysis was conducted on the 18 responses that were provided. All 22 of the control parents completed the item. The mean score on this item for the autism group was 2.72, SD = 1.32, while the mean score on this item for the control group was 2.82, SD = 1.30. The autism group and the control group did not differ on parental ratings using the Grunau et al. pain temperament item, t (38) = -0.231, p > 0.05.

Concordance Between Parental Report and Behavioral Measures

The concordance between parental reports of pain and behavioral measures of pain was tested by examining the correlations between FPS scores of the parents and the facial pain responses and behavioral distress pain responses of the children. For the autism group, no significant correlations were observed between a) the FPS scores provided by the parents and the behavioral distress pain responses of the children, r = -0.12, p > 0.05, nor b) the FPS scores provided by the parents and the facial pain responses of the children, r = -0.15, p > 0.05. A significant correlation was observed between the behavioral distress pain responses and the facial pain responses, r = 0.60, p < 0.01 (see Table 11). For the control group, no significant correlation was observed between FPS scores provided by the parent and the behavioral distress pain responses of the child, r = 0.25, p > 0.05. However, significant correlations were observed between a) FPS scores provided by the parent and facial pain responses of the child, r = 0.61, p < 0.01 and b) behavioral distress pain responses and facial
pain responses of the child, $r = 0.60, p < 0.01$ (see Table 12).

The differences between the above correlations of the autism and control groups were analyzed using the Fisher Z-transformation. The correlations between the FPS scores of the parents and the facial pain responses of the children were significantly different between the autism and control groups, $Z = 2.64, p < 0.01$. However, the correlations between the FPS scores of the parents and the behavioral distress pain responses of the children were not significantly different between the autism and control groups, $Z = 1.15, p > 0.05$.

**Exploratory Analyses**

Exploratory analyses were conducted to examine the concordance between retrospective parental estimates of child pain sensitivity and reactivity and observed behavioral responses. The retrospective parental estimate of child pain sensitivity was the Grunau et al. child pain temperament item while the DPC score served as the retrospective reactivity measure. When completing these items, the parents were asked to think about times when their children had been hurt or in pain. The resulting score is likely due to an amalgamation of multiple observations that the parent had remembered over time and presumably should be related to the behavioral responses of the child.

For the autism group, correlational analyses (see Table 11) showed significant inverse relationships between a) DPC scores and facial pain responses, $r = -0.47, p < 0.05$, b) pain temperament ratings and facial pain responses, $r = -0.50, p < 0.05$, c) DPC scores and behavioral distress pain responses, $r = -0.47, p < 0.05$, and d) pain temperament ratings and behavioral distress pain responses, $r = -0.54, p < 0.05$. Predictably, the retrospective parental measures correlated significantly with one another, $r = 0.75, p < 0.01$. 
The control group showed a different pattern of relationships between the retrospective parental report variables and behavioral observations (see Table 12). Correlational analyses failed to show any significant relationships between a) DPC scores and facial pain responses, $r = 0.32$, $p > 0.05$, b) pain temperament ratings and facial pain responses, $r = 0.36$, $p > 0.05$, c) DPC scores and behavioral distress pain responses, $r = 0.32$, $p > 0.05$, and d) pain temperament ratings and behavioral distress pain responses, $r = 0.13$, $p > 0.05$. However, similar to the autism group, a significant correlation was observed between retrospective parental measures, $r = 0.53$, $p < 0.05$.

Child self-report of pain in the control group did not correlate significantly with any of the behavioral or parental measures (see Table 12).

Discussion

Facial Pain Reactions

The findings of this study clearly go against the prevailing view of the scientific and professional literature which suggests that children with autism are insensitive or indifferent to pain. If the children with autism were insensitive to pain, they should have shown no, or at least diminished, response to the venepuncture procedure. However, as predicted, the children with autism in this study showed increased facial pain reactions in response to a venepuncture and those reactions were comparable to a group of control children without autism. Both groups of children showed similar patterns of low facial activity during baseline with increasing facial activity during the pre-needle and needle phases followed by a decline in facial activity during the post-needle and return to baseline phases. There were no significant differences in overall facial activity between the children with autism and the control children.
throughout the venepuncture procedure. In fact, the only significant difference between the overall facial activity of the two groups was that, compared to the control children, the children with autism displayed greater facial displays during the venepuncture.

The profile of facial activity also differed between the children with autism and the control children; the pain face of the children with autism displayed more facial actions than the control children. The pain face of the children with autism was characterized by brow lower, eye squeeze, flared nostril, nose wrinkle, cheek raise, vertical mouth stretch, and horizontal mouth stretch. The pain face of the control group, in contrast, was characterized by brow lower, squint, and flared nostrils. It is possible that the different topologies of facial displays during the venepuncture are related to cognitive-developmental differences between the two groups of children. The developmental delay and cognitive impairment in the children with autism likely resulted in an automatic facial display relatively independent of higher mental processes. The independence from higher mental mediation likely resulted in less inhibition of facial activity and therefore greater numbers of facial actions were recruited in the pain face. Soussignan, Schaal, Schmit, and Nadel (1995) found that children with pervasive developmental delays displayed more facial actions in response to aversive smells compared to control children. Soussignan et al. (1995) explain that there is a “deficit of socialization of hedonic facial displays in developmentally disordered Ss [children]” (p. 47). In other words, the children with pervasive developmental disorders do not inhibit their facial responses to noxious stimuli because they are not socialized to do so.

When the reactions of the two groups of children were analyzed separately, only the children with autism showed a statistically significant increase in facial activity in response to
the venepuncture. The control children did show a nominal increase in facial activity between
the pre-needle and needle phases, but this increase did not reach levels required for statistical
significance. This was likely due to limited power in detecting differences which resulted from
only having a sample size of 22 children in the control group. With larger sample sizes, the
increase in facial activity between the pre-needle and needle phases would likely reach
significance.

Behavioral Distress Reactions

The results examining the behavioral distress scores for the two groups of children
suggest that the procedure was more distressing for the children with autism than for the
control children. The children with autism had behavioral distress scores greater than the
control group during all phases of the procedure except the baseline phase. This is likely due
to the differences in the procedures themselves rather than differences in the behavioral
reactivity of the children. One of the behaviors coded in OSBD is restraint; all of the children
with autism were restrained during the procedure. The children with autism were required to
be bundled in order to perform the venepuncture and this likely led to an inflation of their
behavioral distress scores. By the nature of the procedure, the children with autism received
scores for restraint during the pre-needle, needle and post-needle phases. In contrast, restraint
was rarely used for the control children at any point in the procedure.

The children with autism and the control children showed the same pattern of
behavioral distress throughout the procedure: low baseline behavioral distress followed by
increasing behavioral distress during the pre-needle and needle phases and then a decline in
behavioral distress during the post-needle and return to baseline phases. Neither group of
children had a significant increase in behavioral distress responses between the pre-needle and needle phases. This is likely a result of OSBD not being specific to the detection of pain. OSBD is designed to detect behavioral distress which includes pain, but also anxiety. Since the procedure was anxiety provoking for both groups of children, they had high behavioral distress scores during the pre-needle phase. This likely resulted in a ceiling effect by which there was little room in the measure to detect pain (above and beyond the anxiety) during the needle phase.

Parental Assessment of Pain Sensitivity and Reactivity

Contrary to expectations, parents' retrospective reports of pain sensitivity and reactivity of their children did not differ between the children with autism and the control children. It was hypothesized that parents of children with autism would report decreased pain sensitivity and reactivity compared to the reports of the parents of the control children. These hypotheses were based on results of a study which reported that parents of children with cognitive impairment perceived decreased pain sensitivity in their children compared to non-impaired children (Fanurik, Koh, Schmitz, Harrison, & Conrad, 1999). However, the parents of the children with autism in the current study were not asked to compare their children to non-impaired children. Rather, the parents were asked to provide ratings for their children without relating those ratings to what they believed about non-impaired children. It is unclear from the results of the current study what the parents of the children with autism know about pain in their children relative to children without autism.

Concordance Between Parental Report and Behavioral Measures

As predicted, the concordance between parental report of pain and facial pain response
of the child was significantly greater for the control group than the autism group. In fact, for the autism group there was no significant correlation between facial activity of the child and parental estimates of pain. It is unlikely that this result can be explained by the parents having difficulty rating their children’s pain with the FPS since the parents of the control children did not seem to have any difficulty doing so.

One possible explanation for the discordance between parental report of pain in the children with autism and the facial reactivity of the child is that the parents of the children with autism were involved participants during the procedure. The parents were required to assist the physicians and nurses in restraining the child during the venepuncture. As a result, the parents may have been distracted from paying attention to the facial displays of their child during the procedure. In contrast, while the parents of the control children were holding their children during the procedure, they were not nearly as active in restraining their children and could have possibly been paying closer attention to facial activity. However, the control parents often had the children sitting on their laps during the procedure which would likely have resulted in the parents not getting a clear view of their child’s face during the entire procedure.

Another possible explanation for the discordance is that there may be some differences in the observations of parents of children with autism when assessing pain in their children. This is not a criticism of parents of children with autism, but a reasonable possibility given the misinformation about pain in children with autism presented in the scientific literature. Reports of pain insensitivity or indifference in children with autism in the literature may be treated as fact by health care professionals who then present the findings to parents. One of
the mothers who participated in the study reported that physicians had told her that her son did not feel pain, but she was convinced he did. Any parents wanting to learn more about autism would likely do their own research on the topic and come across the same literature suggesting pain insensitivity in their children. When viewed in the context of such widespread reporting of pain insensitivity in children with autism, it is not implausible that parents may be uncertain about the pain experience of their children. This could explain the discordance between parental report of pain and facial activity of the children with autism. More research is needed in this area.

Unlike the facial activity results, there was no significant relationship between parental report of pain and behavioral distress pain for either of the groups. This is likely a result of there being no significant increase in behavioral distress between the pre-needle and needle phases for either the children with autism or the control children. This resulted in small behavioral distress pain responses which likely limited the extent of the correlations. As a result, there were no significant differences between the concordance of parental reports of pain and behavioral distress for either of the groups.

Retrospective Parental Judgements of Pain Sensitivity and Observed Pain Behavior

Some of the most perplexing findings emerge from the exploratory analyses which examined the relationship between retrospective parental judgements of pain sensitivity and reactivity in their children and the observed behavioral reactivity of the children in response to the venepuncture. For the control group, the correlations between retrospective parental judgements and observed behavioral reactions of the children were positive, but non-significant. However, for the autism group, there were significant negative correlations
between retrospective parental reports of pain sensitivity/reactivity and observed facial and behavioral distress responses of the children. In other words, the children who were the most reactive during the venepuncture were the ones who were previously identified by their parents as less sensitive and reactive to pain. These results are especially disturbing when considering that the notion of pain insensitivity in children with autism has resulted, in part, from studies which have used retrospective parental reports as the primary measure of pain sensitivity in these children (Gillberg et al., 1985; Gillberg, 1988). The results put into question the validity of using retrospective parental reports to assess pain sensitivity or temperament in children with autism. Again, this may result from confusion on the part of the parents about pain in their children.

There is another possible explanation for the results of discordance between retrospective parental reports and behavioral reactivity of the children with autism. When the parents were asked to complete the DPC and pain temperament item, they were asked to identify a particular painful incident their child had experienced which then served as the basis of their judgments. Many incidents used by parents focused on everyday types of pain (e.g., falling down, bumping into things) and not medical situations. The presentation of pain in children with autism may differ depending on the setting (home vs. hospital) and type of painful incident (everyday pain vs. medical procedures). While the results suggest that children with autism are not insensitive to pain (that is, have an underlying biological deficit which prevents the sensation of pain), context dependant pain indifference cannot entirely be ruled out. It is possible that children with autism show typical behavioral reactions to procedural pain in a clinical setting, but atypical responses to everyday pain in their home.
environment. Future research should explore the concordance between parent report of pain sensitivity and observed reactivity of children with autism to everyday painful incidents.

Results from such research would help clarify whether or not children with autism display context-dependent pain insensitivity.

**Relationship Between Child Self-Report, Parental Report and Behavioral Measures**

One of the most surprising results did not involve the children with autism. The children in the control group provided self-report on how much pain they felt during the venepuncture procedure and their self-report was discordant with all other measures. There is some question about whether the younger children understood the FPS sufficiently enough to use it accurately in providing self-report. As for the older children, different processes may have resulted in the seemingly discordant self-report judgements. Unlike observational measures which tend to capture behavior that is more automatic and less subject to voluntary control, self-report measures capture pain behavior that is under the control of higher mental processes (Hadjistavropoulos & Craig, in press). Many of the children who were observed as being highly reactive to the venepuncture reported feeling little pain on the FPS. Often they would do this with a grin. In addition, most of the children in the control group were boys who were likely raised with the “big boys do not cry” attitude which is prominent in North American culture. These factors put into question the validity of self-report in this particular sample of children.

**A Sociocommunicative Model of Pain in Children with Autism**

The sociocommunicative model of pain which has been proposed elsewhere (Craig, Lilley, & Gilbert, 1996; Hadjistavropoulos & Craig, in press; Prkachin & Craig, 1995) serves
as an excellent model with which to understand pain in children with autism. The model (Figure 3) explains that understanding pain requires an appreciation of the complex social interactions among people in pain and their caregivers (Hadjistavropoulos & Craig, in press). The model begins with a pain stimulus, which in this study was the venepuncture. The painful stimulus of the needle results in an internal experience of nociception in the child. Under the assumption of pain insensitivity in children with autism, this is where the process should stop; pain insensitivity suggests an underlying biophysical change which prevents nociception. This is what the opioid hypothesis aimed at explaining and, to date, that is where the explanation of pain in children with autism has stopped.

However, the results of this study require that the understanding of pain in children with autism needs to be taken further. The children with autism in this study encoded behavioral reactions in response to the pain stimulus. The cognitive and communicative deficits in autism prevent verbal expression of pain and therefore, nonverbal behaviors become the primary source of information about pain. Those behavioral reactions in this study were exhibited as increased facial displays during the needle phase of the procedure. When the pain stimulus ended, the facial displays decreased. The overall pattern of facial displays throughout the procedure was very similar between the children with and without autism.

The final component of the sociocommunicative model of pain is the decoding of the encoded responses of the child by a caregiver. The effectiveness of pain communication relies not only on the encoded displays of the child, but also upon characteristics of the parent (Hadjistavropoulos & Craig, in press). These characteristics include attention and perceptivity as well as an ability to attribute meaning and interpretation to the encoded displays of the child.
Pain in Children with Autism (Craig et al., 1996). In addition to this, cognitive biases about pain in children with autism have an impact on the decoding process. Prkachin and Craig (1995) explain, "The model proposes that each observer's perception of another's pain can be characterized by a gain function which may amplify or attenuate his or her estimate of the evidence about pain coming from the sufferer" (p. 200). In autism, a literature based on anecdotal observations and clinical lore have suggested that children with autism are insensitive to pain. This could potentially bias perceptions of pain in children with autism, reducing the "gain" and attenuating caregiver estimates of pain in these children. The social and communicative deficits inherent in autism further complicate the situation by preventing confirmation of caregiver pain estimates through the child's self-report of pain. In addition, the potential contradiction of observed pain in children with autism and a long-standing belief of pain insensitivity could lead to confusion and difficulty assessing pain. The results of this study support such a possibility.

Therefore, the sociocommunicative model of pain not only serves to explain the complexities of pain in children with autism, but it also provides a mechanism for understanding why children with autism have been viewed as being insensitive to pain. Research in this area needs to focus more on understanding the interplay between pain behaviors of children with autism and the interpretations of those behaviors by caregivers. The simplistic notion of pain insensitivity in children with autism must be abandoned to more fully understand pain in these children.

Future Directions

The current study examined procedural pain in children with autism in the context of a
clinical setting. Future research should explore the responses of children with autism to every day painful events (e.g., falling, bumping into things). Such research would clarify whether or not these children exhibit context-dependent pain indifference. In addition, parental estimates of pain sensitivity in their children should be compared to everyday pain behaviors of the children. It is possible that for everyday pain, parental reports of sensitivity and reactivity of their child may be more concordant with observed behaviors than was found in the present study. As well, future research should address what parents of children with autism believe and understand about the pain experience of their children. This would reveal any confusion or misinformation which may bias parental judgements of pain.

Limitations of the Study

The limitations of the study are similar to many which plague the area of research in autism. The clinical nature of the design resulted in an inability to control all of the variables in the study. For example, there were differences in the venepuncture procedures between the control and autism groups. The procedure for the autism group often required the child to be restrained by a number of adults whereas this was not the case for the control group. As well, there may have been differences in the venepuncture itself. The data for the control group of children were collected at an outpatient blood collection laboratory where phlebotomists draw blood all day, every day. The techniques used by the phlebotomists may have been different from those used by the physicians who performed the venepunctures on the children with autism. Such factors may have resulted in the control children and the autistic children receiving slightly different pain stimuli.

Studies of children with autism are often plagued by small sample sizes and as a result,
limited power in detecting statistically significant findings. While the sample size of 21 children with autism was good compared with other studies in the area, power remains an issue. A number of analyses were identified as likely reaching significance with greater sample sizes (e.g., the increase in facial activity in response to the venepuncture for the control group). The main difficulty in getting a sample of children with autism is that the disorder is relatively rare. Finding children with autism experiencing the same painful stimulus adds even greater difficulty in getting an appropriate sample size for statistical analyses such as correlations. As a result, research in this area needs to be opportunistic, taking advantage of situations as they present themselves. This is the context under which this study took place. The result is less control over the sampling process and a sample size which is limited. There are no easy solutions to this problem; it is just an inevitable consequence of conducting research with children with autism.

Finally, as has already been discussed, the study examined acute pain reactions to a specific medical procedure. Care should be used in generalizing the results of this study to all pain in children with autism. The context and type of pain are important and the results of this study should not be generalized to everyday pain or chronic pain in children. Since the experience of pain is dependant on physical, contextual and social factors, which are constantly changing, there is no single pain experience. Further research is needed in order to determine if the results of this study generalize to different types of pain in children with autism.

Conclusions

The dominant opinion in the scientific literature is that children with autism experience
pain differently than children without autism. Specifically, children with autism have been
viewed as being insensitive or indifferent to pain. The findings of this study question the
validity of such reports. In response to a noxious stimulus, the children with autism in this
study showed a significant behavioral reaction comparable to children without autism. The
unsubstantiated beliefs about pain in children with autism likely have had an impact on
perpetuating the notion of insensitivity by creating confusion and doubt in the minds of
caregivers who are often called upon to provide proxy reports of pain. The
sociocommunicative model of pain takes into account the complexities of the pain experience
in children with autism and helps to explain why seemingly mythical ideas about pain in these
children have been so longstanding. Research has failed to show that children with autism
have altered nociception and the findings of this study show that they do encode pain
responses. The difficulties in understanding pain in these children appear to be in the decoding
of the pain responses by adults. Clinically, these results suggest that children with autism may
be at risk for substandard pain management resulting from an inability to assess their pain
accurately. Valid behavioral and observational measures are needed to assist in the decoding
of the pain responses of children with autism.
References


Table 1
Descriptive Statistics for CFCS Data

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Table 2
Student Newman Keuls Analysis Testing the Differences Between CFCS Segments for the Autism and Control Groups Combined

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* Significant difference at p < 0.05
** Significant difference at p < 0.001
Table 3
Post Hoc t-tests Examining Differences in Facial Activity Scores Between the Autism Group and Control Group Across Segments

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* Significant at p < 0.01
Table 4
Descriptive Statistics for OSBD Data

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Table 5
Student Newman Keuls Analysis Testing the Differences Between OSBD Segments for the Autism and Control Groups Combined

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* Significant difference at p < 0.01  
** Significant difference at p < 0.001
Table 6
Post Hoc t-tests Examining Differences in Behavioral Distress Scores Between the Autism Group and Control Group Across Segments

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* Significant at p < 0.01  
** Significant at p < 0.001
Table 7
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* Significant difference at p < 0.05
** Significant difference at p < 0.001
Table 8
Student Newman Keuls Analysis Testing the Differences in CFCS Scores Between Segments for the Control Group

<table>
<thead>
<tr>
<th></th>
<th>Pre-Needle</th>
<th>Needle</th>
<th>Post-Needle</th>
<th>Return to Baseline</th>
</tr>
</thead>
<tbody>
<tr>
<td>Baseline</td>
<td>1.34</td>
<td>3.13**</td>
<td>0.59</td>
<td>0.1</td>
</tr>
<tr>
<td>Pre-Needle</td>
<td></td>
<td>1.80^A</td>
<td>0.75</td>
<td>1.43</td>
</tr>
<tr>
<td>Needle</td>
<td></td>
<td></td>
<td>2.54*</td>
<td>3.23**</td>
</tr>
<tr>
<td>Post-Needle</td>
<td></td>
<td></td>
<td></td>
<td>0.69</td>
</tr>
</tbody>
</table>

^ Significant difference at p < 0.10
* Significant difference at p < 0.05
** Significant difference at p < 0.01
Table 9
Student Newman Keuls Analysis Testing the Differences in OSBD Scores Between Segments for the Autism Group

<table>
<thead>
<tr>
<th></th>
<th>Pre-Needle</th>
<th>Needle</th>
<th>Post-Needle</th>
<th>Return to Baseline</th>
</tr>
</thead>
<tbody>
<tr>
<td>Baseline</td>
<td>1.92*</td>
<td>2.16*</td>
<td>1.70*</td>
<td>0.87*</td>
</tr>
<tr>
<td>Pre-Needle</td>
<td>0.24</td>
<td>0.22</td>
<td>1.06*</td>
<td></td>
</tr>
<tr>
<td>Needle</td>
<td></td>
<td>0.46</td>
<td>1.29*</td>
<td></td>
</tr>
<tr>
<td>Post-Needle</td>
<td></td>
<td></td>
<td>0.83*</td>
<td></td>
</tr>
</tbody>
</table>

* Significant difference at p < 0.001
Table 10
Student Newman Keuls Analysis Testing the Differences in OSBD Scores Between Segments for the Control Group

<table>
<thead>
<tr>
<th></th>
<th>Pre-Needle</th>
<th>Needle</th>
<th>Post-Needle</th>
<th>Return to Baseline</th>
</tr>
</thead>
<tbody>
<tr>
<td>Baseline</td>
<td>0.55</td>
<td>0.91*</td>
<td>0.18</td>
<td>0.05</td>
</tr>
<tr>
<td>Pre-Needle</td>
<td></td>
<td>0.36</td>
<td>0.36</td>
<td>0.59</td>
</tr>
<tr>
<td>Needle</td>
<td></td>
<td></td>
<td>0.73*</td>
<td>0.95*</td>
</tr>
<tr>
<td>Post-Needle</td>
<td></td>
<td></td>
<td></td>
<td>0.23</td>
</tr>
</tbody>
</table>

* Significant difference at p < 0.05
Table 11
Correlation Table for Autism Group

<table>
<thead>
<tr>
<th></th>
<th>DPC</th>
<th>Parent FPS Rating</th>
<th>Facial Pain Response</th>
<th>Behavioral Distress Pain Response</th>
</tr>
</thead>
<tbody>
<tr>
<td>Pain Temperament</td>
<td>0.75**</td>
<td>0.13</td>
<td>-0.50*</td>
<td>-0.54*</td>
</tr>
<tr>
<td>DPC</td>
<td>-0.03</td>
<td>-0.47*</td>
<td>-0.47*</td>
<td></td>
</tr>
<tr>
<td>Parent FPS Rating</td>
<td></td>
<td>-0.15</td>
<td></td>
<td>-0.12</td>
</tr>
<tr>
<td>Facial Pain Response</td>
<td></td>
<td></td>
<td></td>
<td>0.60**</td>
</tr>
</tbody>
</table>

* Significant at p < 0.05 (2-tailed)
** Significant at p < 0.01 (2-tailed)
Table 12
Correlation Table for Control Group

<table>
<thead>
<tr>
<th></th>
<th>Parent FPS Rating</th>
<th>Pain Temperament</th>
<th>DPC</th>
<th>Facial Pain Response</th>
<th>Behavioral Distress Pain Response</th>
</tr>
</thead>
<tbody>
<tr>
<td>Child FPS Rating</td>
<td>0.28</td>
<td>0.12</td>
<td>-0.06</td>
<td>0.25</td>
<td>0.29</td>
</tr>
<tr>
<td>Parent FPS Rating</td>
<td></td>
<td>0.28</td>
<td>0.22</td>
<td>0.61**</td>
<td>0.25</td>
</tr>
<tr>
<td>Pain Temperament</td>
<td></td>
<td></td>
<td></td>
<td>0.53*</td>
<td>0.36</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>0.32</td>
<td>0.32</td>
<td></td>
</tr>
<tr>
<td>Facial Pain Response</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>0.60**</td>
</tr>
</tbody>
</table>

* Significant at p < 0.05 (2-tailed)
** Significant at p < 0.01 (2-tailed)
Pain in Children with Autism
Figure 2: Graph of behavioral distress across coding segments
Figure 3: Sociocommunicative model of pain (adapted from Craig et al., 1996)
APPENDIX A: Faces Pain Scale
PAIN QUESTIONNAIRE
(please complete this questionnaire and bring it to your next study appointment)

When answering the following questions, please think about times when your child has been hurt or in pain. For example, think about times when you have seen something happen to your child that would be painful for most people, or have seen actual tissue damage (like a burn or a cut), or if you knew for sure your child had a painful condition (like an earache that has been verified by a physician).

A. Rate the following item for your child on a scale from 1 (not characteristic or typical of your child) to 5 (very characteristic of your child).

1. My child is very sensitive to pain of bumps or cuts or other common hurts.

<table>
<thead>
<tr>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
</tr>
</thead>
<tbody>
<tr>
<td>not typical/characteristic</td>
<td>very typical/characteristic</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

B. Please indicate how often the following behaviours occur when your child is hurt or in pain.

1. Moaning, whining, whimpering (fairly soft) 1 2 3 4
2. Crying (moderately loud) 1 2 3 4
3. Screaming/yelling (very loud) 1 2 3 4
4. A specific sound or word for pain (e.g., a word, cry, or type of laugh) 1 2 3 4
5. Eating less, not interested in food 1 2 3 4
6. Increase in sleep 1 2 3 4
7. Decrease in sleep 1 2 3 4
8. Not co-operating, cranky, irritable, unhappy 1 2 3 4
9. Less interaction with others, withdrawn 1 2 3 4
10. Seeking comfort or physical closeness 1 2 3 4
11. Being difficult to distract, not able to satisfy or pacify 1 2 3 4
12. A furrowed brow 1 2 3 4
13. A change in eyes, including: squinting of eyes, eyes opened wide, eyes frowning 1 2 3 4
14. Turning down of mouth, not smiling 1 2 3 4
15. Lips puckering up, tight, pouting or quivering 1 2 3 4
16. Clenching or grinding teeth, chewing or thrusting tongue out
17. Not moving, less active, quiet
18. Jumping around, agitated, fidgety
19. Floppy
20. Stiff, spastic, tense, rigid
21. Gesturing to or touching part of the body that hurts
22. Protecting, favouring, or guarding part of the body that hurts
23. Flinching or moving the body part away, being sensitive to touch
24. Moving the body in specific ways to show pain (e.g., head back, arms down, curls up, etc.)
25. Shivering
26. Change in colour, pallor
27. Sweating, perspiring
28. Tears
29. Sharp intake of breath, gasping
30. Breath holding

Please indicate what type(s) of hurt(s) or pain(s) you were thinking about when completing this questionnaire.
APPENDIX C: Brief cognitive screen for the control group

Pain in Children Participant Information

Child’s Name: ________________________________ # ______
Date of Procedure: ________________________________

1. Child’s Date of Birth (dd/mm/yy): ________________________________
2. Child’s Gender (circle): M □ F □
3. Has your child ever had a central nervous system disorder? Yes □ No □
4. Has your child ever had a serious head injury? Yes □ No □
5. Has your child ever experienced oxygen deprivation? Yes □ No □
6. Have you ever suspected a developmental delay in your child? Yes □ No □
7. Have your child’s teachers/daycare providers ever expressed concern about your child’s development or possible learning difficulties? Yes □ No □
8. Will your child be undergoing a formal cognitive assessment anytime in the next 6 months? Yes □ No □
9. Is your child currently on any pain medication? Yes □ No □
10. Has your child worn an EMLA patch today? Yes □ No □

If you answered “Yes” to any of the above questions, please explain:

_________________________________________________________________
_________________________________________________________________
_________________________________________________________________
_________________________________________________________________
_________________________________________________________________
_________________________________________________________________
_________________________________________________________________
_________________________________________________________________
concerns about treatment as a research subject can be referred to Dr. R.D. Spratley, Director, Office of Research Services, UBC, 822-8598.

Parent Consent: I understand that the participation of my child in the above study is entirely voluntary and that I may refuse to participate or may withdraw my child from the study at any time without any consequences to my child's involvement with Sunny Hill Health Centre for Children. In particular, I understand that if I chose not to participate in this study, my child will still be able to participate in the Secretin Treatment Study. I have received a copy of this consent form for my own records. I consent to the participation of my child in this study.

Parent's/Legal Guardian's signature

Date

Witness's signature
Contacts: The investigators would be happy to answer any questions at any time regarding this study to ensure that the participants understand completely what it involves. Any concerns about treatment as a research subject can be referred to Dr R.D. Spratley, Director, Office of Research Services, UBC, 822-8598.

Parent Consent: I understand that the participation of my child in the above study is entirely voluntary and that I may refuse to participate or may withdraw my child from the study at any time without any consequences to my child's involvement with BC Children's Hospital. I have received a copy of this consent form for my own records. I consent to the participation of my child in this study.

Parent's/Legal Guardian's signature

Date

Witness's signature