

Expression of Pain in Children With Autism

Rami Nader, MA,* Tim F. Oberlander, MD, FRCPC,† Christine T. Chambers, PhD,‡§ and Kenneth D. Craig, PhD*

Objectives: Reduced pain sensitivity is widely reported to be a common feature of children with autism, yet this conclusion frequently has been based on anecdotal observations and questionable measures of pain. The aims of the study were to (1) characterize the behavioral response of children with autism experiencing a venepuncture using objective observational measures of pain and distress, (2) examine parents' assessments of pain behavior in children with and without autism, including comparison of the relationship of parental reports with behavioral measures, and (3) compare the behavioral reactions and parental assessments of children with autism with children without autism undergoing venepuncture.

Methods: Pain reactions to the invasive procedure of venepuncture were videotaped, systematically described and compared in 21 children with autism (3–7 years old) and 22 nonimpaired children, the latter providing a chronological age and gender equivalent comparison group. Parents provided observer reports of pain, and facial activity was used as an objective behavioral measure of pain.

Results: The children with autism displayed a significant facial pain reaction in response to the venepuncture procedure. There was a lack of concordance between parental reports of pain and observed pain responses for the children with autism. Behavioral responses of the children with autism were generally similar to the comparison group, except the substantial facial pain reactivity instigated by the vene-

puncture in the children with autism exceeded that displayed by the nonimpaired comparison children. Parent reports of pain severity did not differ between the autism and comparison groups. The degree of concordance between parental report and observed pain responses was consistently better for the comparison group.

Discussion: The findings demonstrate that children with autism display a significant behavioral reaction in response to a painful stimulus, and these findings are in sharp contrast to the prevailing beliefs of pain insensitivity described in the literature to date. The findings also raise questions about the appropriateness of parental global report as an assessment tool for pain in children with autism.

Key Words: autism, pain, facial expression, behavioral measures, parent report

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Despite a vast body of research on autism and popular media fascination with this disorder, relatively little empirical knowledge is available regarding pain among children with autism. Children with autism have been described as having “reduced pain sensitivity”,^{1–4} “not feeling pain as intensely as others”,⁵ having an “indifference to pain”,⁶ and having a “high threshold for pain”.⁷ Remarkably, most of these reports of altered pain sensation have been based on anecdotal observations and clinical impressions.^{6,8} These generalizations have not been examined in detail, and adequate empirical documentation has not been provided to support these assertions.

Autism is a childhood developmental disorder with a reported prevalence of about 5 cases per 10,000,⁷ although this estimate may be lower than the true prevalence of the disorder.⁹ Autism is characterized by deficits in behavior, social interaction, and communication. Children with autism frequently have abnormal social relationships and deficits in verbal and nonverbal communication.⁹ This is further compounded by stereotypic and restricted patterns of behavior, interests, and activities.⁷ Autism is also associated with cognitive impairment (75% of children with autism function at an intellectually disabled level).¹⁰

To date, the primary focus of research on pain in children with autism has been to explain a possible mechanism underlying presumed reduced pain sensitivity, in particular, a hypothesized hyperfunctioning endogenous opioid system.^{1,3–5,11}

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*From the Department of Psychology, University of British Columbia, Vancouver, BC, Canada; †Division of Developmental Pediatrics, Sunny Hill Health Centre for Children, and Centre for Community Child Health Research, University of British Columbia, Vancouver, BC, Canada; ‡Department of Pediatrics, University of British Columbia, Vancouver, BC, Canada; and §Centre for Community Child Health Research, BC Research Institute for Children's and Women's Health, Vancouver, BC, Canada.

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Reprints: Rami Nader, MA, Department of Psychology, University of British Columbia, 2136 West Mall, Vancouver, BC, Canada V6T 1Z4 (e-mail: mader@interchange.ubc.ca).

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The opioid hypothesis ostensibly accounts for many symptoms associated with autism, including pain insensitivity.³ Several mechanisms have been proposed to account for opioid anomalies.³ Endogenous opioid levels may be increased for genetic reasons. These could cause primary overproduction, deficient degradation, abnormal messenger mechanisms within the nerve cells, or feedback deregulation mechanisms. Another hypothesis has been that repetitive stereotypic motor behaviors (including self-injury), commonly observed in children with autism, could increase brain opioid levels, indirectly producing euphoria, some autistic symptoms, and pain insensitivity.³

Research findings addressing supposed opioid systems in children with autism have been mixed.³ Early studies^{1,5} supported the hypothesis but suffered from methodological problems. Control groups were not equivalent to the autism groups in terms of age or sex ratio, and investigators did not use validated approaches to assessing pain sensitivity or prospective data (their prime source was retrospective parental report). Proxy reports of pain in children without autism frequently pose problems as they can be unreliable and most often underestimate the child's self-report of pain when that is available.¹²

An alternative explanation for the apparent pain insensitivity in children with autism derives from a sociocommunicative perspective.¹³ Inadequate communication skills and deficiencies in social relatedness consistently distinguish children with autism from other children.¹⁴ Kanner, who first documented the disorder, regarded social dysfunction and unusual responses to the social environment as 2 essential features of autism.¹⁵

The impairments found in children with autism traditionally would be considered left hemisphere mediated. Verbal, sequential processing, and analytic skills are all almost uniformly poor or absent in children with autism.^{15,16} Skills mediated by the right hemisphere (eg, visuospatial skills) are usually less impaired.¹⁶ A delay or total lack of development of spoken language also characterizes this population.^{7,17} If speech does develop, it tends to be atonal, lacks inflection, and fails to convey emotions.¹⁰ When available, their language is characterized by concreteness, repetitiveness, and a mechanical/noncommunicative approach.¹⁶

Autism is also characterized by impairment in certain nonverbal and social behaviors, including a failure to develop social relationships.¹⁷ 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.^{7,10,17} They prefer to be alone and seem to ignore humans while attending to nonhuman objects in the environment.^{10,18}

Given this lack of social responsiveness and language impairment, the expression of pain would also likely be altered, contributing to perceptions of atypical pain experience by parents or other caregivers. 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 inanimate objects. Even if the children were to approach their parents, they would have difficulty verbally communicating their pain experience. They also have difficulties in the use of body gestures and understanding the language of others.¹⁷ Therefore, it is not surprising that these children do not seem to express pain or readily seek comfort from others when in distress.

Parents of children with cognitive or neurologic impairments leading to communication limitations frequently rely upon nonverbal behavior to determine if their child is in pain.^{19,20} It is noteworthy that a majority of parents of children with cognitive impairment believe their children experience pain differently than children without cognitive impairment.²⁰ Parents of children with cognitive impairment tend to perceive decreased pain sensitivity and increased pain tolerance in their children compared with other nonimpaired children.²⁰ These cognitive expectancies may further increase the likelihood of parental underestimation of pain in children with cognitive impairment. The questionable validity of proxy reports demands contrast with objective measures, as in recent studies.^{21–23}

The present study had several objectives: (1) To characterize the behavioral response of children with autism experiencing an invasive medical procedure (venepuncture) using objective observational measures of pain and distress. (2) To examine parents' assessments of pain behavior in children with autism, including comparison of the relationship of parental reports with behavioral measures, as well as carry out exploratory analyses to examine the relationship between retrospective parental reports of pain sensitivity and reactivity in children and observed expressions of pain in children during a venepuncture. (3) To conduct a preliminary comparison of pain behavior and parental assessment of pain in children with autism and nonimpaired children experiencing similar painful procedures.

MATERIALS AND METHODS

Participants

Twenty-one children with autism were recruited at a health center serving children with developmental delays. The sample of children with autism consisted of 18 boys and 3 girls with a mean age of 5.42 years (SD = 1.13 years, range 3.17–7.00 years). The children and their parents were participating in a study²⁴ examining the effects of a hormone treatment administered via an intravenous injection, using a small gauge (25 Gx 3/4") butterfly needle. Inclusion criteria for children with autism included (a) a score of 30 or more on the Childhood Autism Rating Scale (CARS)²⁵; (b) a score of 6 or more on the DSM-IV diagnostic criteria; and (c) clinical judgment by a pediatrician, psychiatrist, or registered psychologist experienced in the field of pervasive developmental disorders.

Nineteen of the children had Vineland Adaptive Behavior Composite Scores ($M = 45.74$, $SD = 7.75$). The mean CARS score for the autism group was 39.10 ($SD = 4.98$, range 30.5–47), which put the average for the group into the severely autistic range (CARS score > 37).²⁵ Nine children fell into the mildly–moderately autistic range (CARS score 30–37), and 12 children fell into the severely autistic range.²⁵ The parents of the children with autism also completed the Autism Behavior Checklist (ABC).²⁶ The mean ABC total score was 76.3 ($SD = 19.2$, range 38–121). The ABC also breaks down into 5 symptom areas: sensory, relating, body and object use, language, and social. The ABC scores for the subscales were as follows: 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 were similar to norms reported for children with autism.²⁶

A nonimpaired comparison group was also recruited through an outpatient blood collection laboratory at an acute-care children's hospital. The children in the comparison group received a venepuncture (for the purposes of blood collection) similar to the children with autism group (a similar gauge butterfly needle was used), and this allowed for comparisons between the groups since both experienced a similar pain stimulus. The comparison 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 between the ages of 3 and 8 years were recruited in a sex ratio proportional to the autism group. The children in the comparison group were equated to the children in the autism group on the basis of gender ratio and chronological age using a frequency distribution control technique.²⁷ The children were screened for cognitive impairment by having parents complete a short questionnaire concerning the child's relevant medical history (presence of central nervous system disorder, history of head injury or anoxia) and questions asking if teachers, daycare workers, or parents themselves had identified or suspected developmental delays.²⁸ Children experiencing any of these were excluded. Children currently on analgesic medication also were ineligible for participation. All parents and children were English speaking.

Measures

Child Facial Coding System (CFCS)

The CFCS⁴² is a facial coding system designed to assess children's pain experiences. Using explicit criteria, a trained CFCS coder identifies the frequency and intensity of 13 facial actions using stop-frame and slow-motion video editing equipment. CFCS has been used to assess both acute²² and persistent³⁰ pain in children. Facial activity has been identified as a sensitive and relatively specific index of pain in children,³¹ people with communication limitations,³² and people with significant neurologic impairment.²² CFCS scores for each facial action per coding segment were obtained by calculating the

average of the individual facial 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.

Observational Scale of Behavioral Distress (OSBD)

The OSBD^{33,34} is a behavioral coding system designed to assess behavioral distress in children undergoing painful medical procedures. OSBD consists of 8 operationally defined behaviors indicative of anxiety and/or pain behavior in children.³⁴ It is a reliable and valid measure of children's distress in medical situations such as venepuncture and injections.^{33,35,36} OSBD scores for each distress behavior per segment were obtained in a similar fashion as CFCS scores. The average of the individual distress behavior scores for each segment was calculated. In keeping with previous research, a composite behavioral distress score for each segment was calculated by summing the average scores for each of the distress behaviors in each segment.^{34,37}

Faces Pain Scale (FPS)

Parental report of pain was measured using the FPS.³⁸ The FPS consists of 7 faces showing gradual increases in pain expression from left to right (neutral to pain). The FPS is widely used in research and clinical practice and has been used to provide observer measures of pain.^{39,40}

Retrospective Parental Report of Pain Sensitivity and Reactivity

Histories of sensitivity to pain were assessed by having the parents report on prior pain reactions of their children using the Non-Communicating Children's Pain Checklist (NCCPC).¹⁹ This is a 30-item checklist of behaviors caregivers use to determine if people with cognitive impairment are in pain. This scale has been shown to be a valid and reliable measure of pain in this population.⁴¹ The items comprising the checklist are similar to those in behavioral measures used with nonimpaired populations,⁴² and therefore, the checklist was used with both groups to facilitate comparisons. When completing the items on the NCCPC, parents were asked to think about occasions when their children had been hurt or in pain. The resulting score represented an amalgamation of multiple observations that the parent had remembered over time and believed were related to the behavioral responses of the child. Parents also provided a summary 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".⁴³ The parent responded to this question on a scale of 1 = not typical/characteristic to 5 = very typical/characteristic.

Procedure

The study was approved by the University of British Columbia Research Ethics Board and the Children's and Wom-

en's Health Centre of British Columbia Research Review Committee. Parental consent was obtained. For the children with autism, the investigators of the hormone study had sent the families a letter describing the study and the NCCPC to complete beforehand. If interested in participating, they were to mail the signed informed consent form and return the completed NCCPC on the day of the procedure. Additional demographic and psychologic testing information was collected in a chart review of consenting families.

On the day of the injection, the children were videotaped for a baseline period (approximately 3–5 minutes) before the injection procedure began. The baseline video was segmented into 10-second segments and a randomly selected 10-second segment from this period served as the “baseline” phase for coding. As the physician prepared the injection site, nurses and staff wrapped the child in a blanket for the purpose of restraining the child for the procedure. This visibly produced a state of anticipatory distress in the child, thereby providing a nonnoxious, but aversive, control to contrast with the reaction to the venepuncture itself. The 10 seconds immediately preceding the injection served as the “pre-needle” phase for coding. The 10 seconds immediately after the needle insertion was the “needle” phase. Once the needle was inserted and in place, the procedure took approximately 1–2 minutes, after which the needle was removed. The 10 seconds immediately after the needle was removed provided the “post-needle” phase. After the child was unbundled and free to move, the researcher continued to videotape the child for a period of time (approximately 3–4 minutes) from which a randomly selected 10-second segment was chosen as the “recovery” phase. All of the 10-second segments were discrete and did not overlap with any other segments. When the procedure was finished, 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.

For the comparison group, parents were approached upon arrival at the blood collection laboratory. The study was explained, and the parents were given an information letter and informed consent form. If they agreed to participate, and no developmental delays were reported, the parents were given the NCCPC to complete as they waited for their child to be called into the procedure room. Videotaping was conducted in parallel with the procedure for the autism group. Bundling was not required, but, consistent with previous research,⁴⁴ the pre-needle anticipation phase still had the potential to be aversive. The procedure required approximately 1 minute, with parents then rating the child's pain using the FPS. The FPS was completed by the mother in 16 cases and by the father in 6 cases.

Videotapes were CFCS coded for the 5 selected segments (baseline, pre-needle, needle, post-needle, recovery) by trained coders who had achieved high reliability in training. All 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. For OSBD coding, the entire

10-second segment was coded as a single unit, resulting in one score for each OSBD behavior per segment. Since all of the children with autism were bundled for the procedure, 2 of the scored OSBD behaviors (flail and restraint) were not included in the analyses. The reliability of coders was checked by having one coder code all of the segments and another coder code 20% of those segments randomly selected.

Data analysis

For the CFCS data, a 5-level, one-way analysis of variance (ANOVA) with repeated measures was used to determine if there were significant differences in overall facial activity across any of the segments for the autism group. Student–Newman–Keuls post hoc tests were conducted, when appropriate, to determine where differences between segments were significant. When comparing the autism with the comparison group, a 2 (group: autism versus comparison) \times 5 (coding segment: baseline, pre-needle, needle, post-needle, and recovery) ANOVA with repeated measures was used to determine if there were significant differences in facial activity across any of the segments and between the autism and comparison groups as well as interactions. In the case of a significant interaction, simple effects were examined. Post hoc *t* tests (two-tailed) were used to detect simple effects between the groups. Similar analyses were used for the OSBD data.

RESULTS

Coding Reliability

Inter-rater scoring reliability for the CFCS and OSBD data was calculated using the formula recommended by Ekman and Friesen,⁴⁵ which assesses the proportion of agreement on actions recorded by 2 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 comparison group CFCS coding was 0.78, levels similar to other studies⁴⁶ and deemed acceptable. The inter-rater reliability for the OSBD coding of the autism group was 0.84, while the reliability for the comparison group OSBD coding was 0.88.

CFCS and OSBD Analyses

As depicted in Figure 1, the overall facial activity of the children with autism varied across segments during the procedure, $F(4, 80) = 25.75, P < 0.001$. Relative to baseline, the greatest facial activity was observed during the needle segment ($P < 0.001$), followed by the pre-needle ($P < 0.001$) and post-needle ($P < 0.01$) segments. There was a significant increase in facial activity between the pre-needle and needle segments ($P < 0.01$).

As depicted in Figure 2, the overall behavioral distress of the children with autism varied across segments during the procedure, $F(4, 80) = 15.23, P < 0.001$. Relative to baseline, the greatest facial activity was observed during the needle seg-

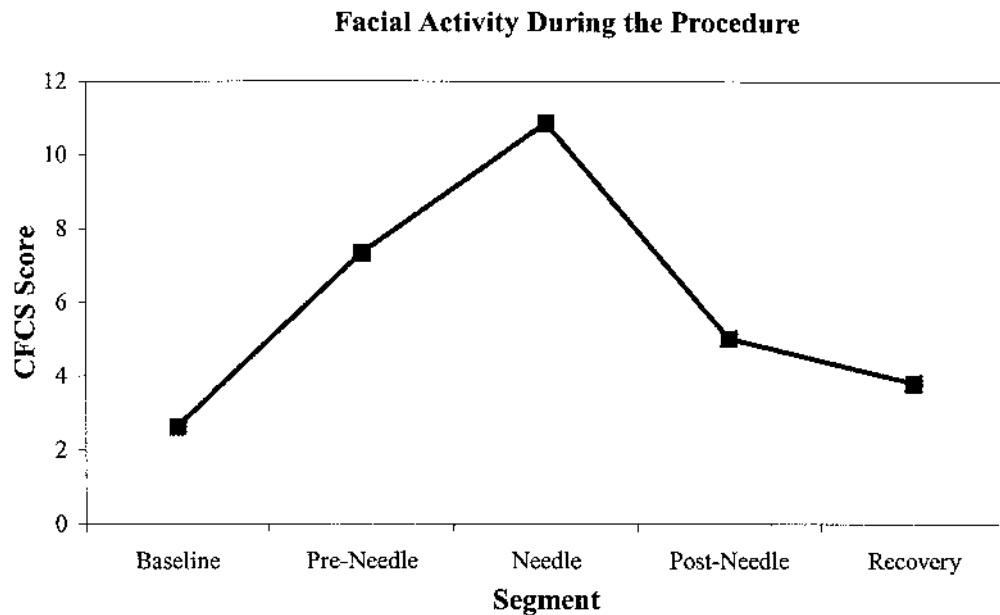


FIGURE 1. Graphic description of facial activity for the children with autism during the procedure.

ment ($P < 0.001$), followed by the pre-needle ($P < 0.001$), post-needle ($P < 0.001$), and recovery ($P < 0.001$). However, unlike facial activity, there was no significant difference in behavioral distress between the needle and pre-needle segments ($P > 0.05$).

Relationship Between Parental Report and Behavioral Measures

Concordance between parental reports of pain and behavioral measures of pain was examined using correlations be-

tween FPS scores of the parents and the facial pain responses of the children. For the autism group, no significant correlation was observed between the FPS scores provided by the parents and the facial pain responses of the children, $r = -0.154$, $P > 0.05$.

Exploratory analyses examined concordance between retrospective parental estimates of child pain sensitivity and reactivity and observed behavioral responses. Children with autism who had been assessed by their parents as having lower pain sensitivity and reactivity tended to show greater facial

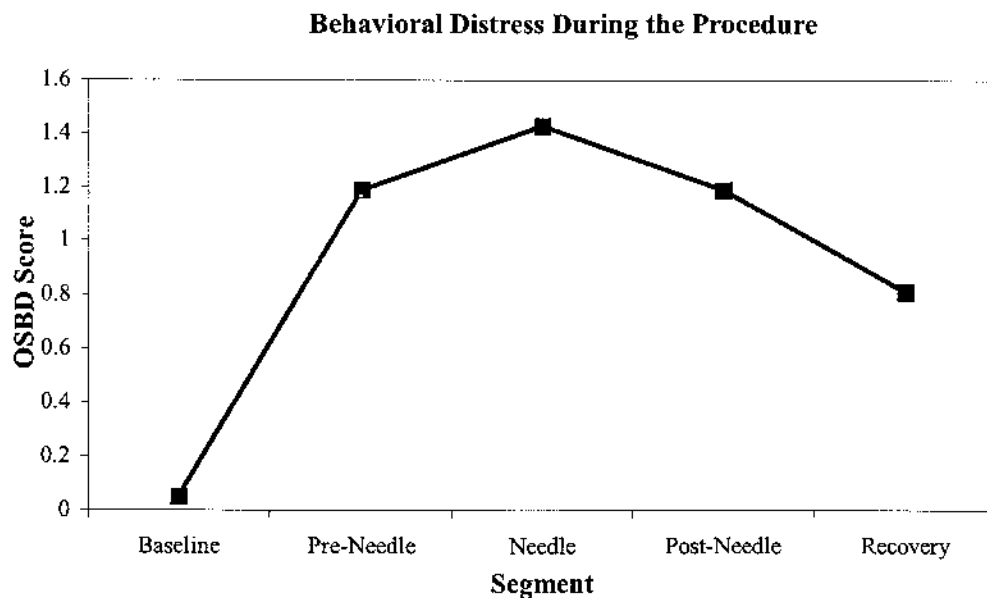


FIGURE 2. Graphic description of behavioral distress for the children with autism during the procedure.

reactions and behavioral distress in response to the venepuncture. For the autism group, correlational analyses showed significant inverse relationships between (a) NCCPC scores and facial pain responses [$r = -0.468, P < 0.05$], (b) pain temperament ratings and facial pain responses [$r = -0.495, P < 0.05$], (c) NCCPC scores and behavioral distress pain responses [$r = -0.469, P < 0.05$], and (d) pain temperament ratings and behavioral distress pain responses [$r = -0.541, P < 0.05$]. Predictably, the retrospective parental measures correlated significantly with one another [$r = 0.750, P < 0.01$].

Comparison Between Autism and Comparison Group

As seen in Table 1, the autism and comparison groups followed a similar general trend of increasing facial activity through the baseline, pre-needle, and needle phases, followed by a decline in facial activity during the post-needle and recovery phases, $F(4, 164) = 24.64, P < 0.001$. Relative to baseline, the greatest facial activity was observed during the needle segment ($P < 0.05$) followed by the pre-needle segment ($P < 0.05$).

There was no significant main effect difference in facial expressions between the groups across the course of the procedure, $F(1, 41) = 2.75, P > 0.05$; however, a significant interaction between groups and segments was observed, $F(4, 164) = 4.59, P < 0.01$. Tests for simple effects during each segment using post hoc *t* tests indicated the autism group had significantly greater facial activity during the needle phase than the comparison group, $t(41) = 3.16, P < 0.01$. There were no other significant differences between the autism and comparison groups during the other segments ($P > 0.05$).

The 2 groups together showed increasing and decreasing behavioral distress throughout the procedure, $F(4, 164) = 19.12, P < 0.001$ (Table 1), similar to the facial activity. However, unlike the CFCS results, there was no significant difference in behavioral distress between the needle and preneedle segments ($P > 0.05$).

Observed behavioral distress in response to the procedure differed between the autism and comparison groups, $F(1, 41) = 4.69, P < 0.05$, with the autism group being more distressed by the procedure. In addition, there was a significant interaction between the groups and segments, $F(4, 164) = 3.36, P < 0.05$. Simple effects analyses indicated that the autism group displayed greater behavioral distress during the post-

needle segment [$t(41) = 3.45, P < 0.01$] and recovery segment [$t(33.9) = 2.12, P < 0.05$] but not during baseline, preneedle, or needle segments ($P > 0.05$).

Using FPS scores as a measure of parental assessment of pain response following the venepuncture, parents of children with autism reported observing more pain in their children during the venepuncture ($M = 4.29, SD = 1.45$) compared with parents of the children without autism ($M = 2.75, SD = 1.90$; $t(41) = 2.97, P < 0.05$). Using the NCCPC as a retrospective measure of parental assessment of typical pain reactivity in their children, scores did not differ between the autism group ($M = 60.33, SD = 13.50$) and comparison group ($M = 58.41, SD = 14.19$; $t(41) = 0.46, P > 0.05$).

Parent reports of pain temperament in children with autism ($M = 2.72, SD = 1.32$) were similar to parent reports of pain temperament in the children without autism ($M = 2.82, SD = 1.30$; $t(38) = -0.23, P > 0.05$). For the autism group, 3 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 comparison parents completed the item.

In contrast to the children with autism, for the children without autism, a significant correlation was observed between FPS scores provided by the parent and facial pain responses of the child [$r = 0.612, P < 0.01$]. The relationship between parental assessment of pain and observed behavioral reactivity of the children in response to the venepuncture was greater for the comparison group compared with the autism group [$Z = 2.64, P < 0.01$].

In addition, the comparison group showed a different pattern of relationships between the retrospective parental report variables and behavioral observations. Correlational analyses failed to show any significant relationships between (a) NCCPC scores and facial pain responses [$r = 0.320, P > 0.05$], (b) pain temperament ratings and facial pain responses [$r = 0.358, P > 0.05$], (c) NCCPC scores and behavioral distress pain responses [$r = 0.247, P > 0.05$], and (d) pain temperament ratings and behavioral distress pain responses [$r = 0.101, P > 0.05$]. However, similar to the autism group, a significant correlation was observed between retrospective parental measures [$r = 0.527, P < 0.05$].

DISCUSSION

This study found that children with autism display a substantial facial pain reaction in response to a venepuncture pro-

TABLE 1. Means and Standard Deviations for CFCS and OSBD Data for the Autism and Comparison Groups

		Baseline	Preneedle	Needle	Postneedle	Recovery
CFCS scores	Autism	2.64 (2.15)	7.34 (4.29)	10.87 (3.72)	5.02 (3.37)	3.81 (2.62)
	Comparison	3.82 (3.39)	5.15 (4.07)	6.95 (4.38)	4.41 (3.18)	3.72 (3.12)
OSBD scores	Autism	0.05 (0.23)	1.19 (0.93)	1.43 (0.87)	1.19 (0.87)	0.81 (0.98)
	Comparison	0.23 (0.75)	0.77 (1.02)	1.04 (0.95)	0.41 (0.59)	0.27 (0.63)

cedure. This finding stands in contrast to the prevailing view in the scientific and professional literature, which suggests that children with autism may be insensitive or indifferent to pain. The outcome contradicts suppositions of atypical endogenous opioid mechanisms suppressing pain reactivity in children with autism. The children with autism displayed an increase in the facial display of pain during the needle segment that was greater than the facial activity during the pre-needle segment. It could be suggested that the facial activity was in response to the bundling procedure and not the venepuncture. Both the facial coding and OSBD data indicated that bundling led to increased behavioral distress in the child prior to the venepuncture. However, facial activity increased even further during the needle segment. The only procedural difference between those segments was the venepuncture itself. Therefore, it is reasonable to conclude that the increase in facial activity was caused by pain resulting from the venepuncture.

The children with autism did not display a significant increase in OSBD behavioral distress responses between the pre-needle and needle phases. This was likely a result of OSBD not being specific to the detection of pain. OSBD was designed to detect behavioral distress, which includes not only pain but also anxiety. Since the procedure was likely anxiety provoking for the children with autism, they had high behavioral distress scores during the pre-needle phase. There likely was a ceiling effect, leaving little room for the measure to detect pain (above and beyond the anxiety) during the needle phase.

For the children with autism, there was no relationship between facial activity of the child and parental estimates of pain. The reason for this finding is not readily clear. The FPS has been used with ease to provide observer measures of pain in other studies,^{39,40} and parents of the children without autism in the current study successfully provided ratings, so it is unlikely that the parents of children with autism would ordinarily have had difficulty using the FPS. Conceivably, the difficulty reflects the challenges all adults encounter understanding the psychologic status of children with autism. Health professionals frequently report pain assessment in children with developmental delays to be difficult, so it is not surprising that parents also might have difficulty in this area. In addition, children with developmental disabilities often have idiosyncratic behaviors like vocal abnormalities or facial peculiarities in response to pain.²⁰ These behaviors could increase the ambiguity of pain-producing situations for parents and health care providers of children with developmental disabilities. This could lead to greater difficulty in providing pain assessments. An alternative explanation is that the idiosyncratic behaviors of the children with autism may have inflated the behavioral pain scores. It is possible that parents took these particular behaviors into account and made judgments regarding their meaning depending on the context. Therefore, it is unclear whether parents take idiosyncratic behaviors into account when making

their judgments or if the idiosyncratic behaviors lead to confusion and uncertainty in the judgments. Further research is needed to clarify this issue.

The most perplexing findings emerged from the exploratory analyses, which examined the relationship between retrospective parental judgments of pain and the observed behavioral reactivity of the children during the venepuncture. For the autism group, there were strong significant negative correlations between retrospective parental reports of pain sensitivity/reactivity and observed behavioral responses of the children. The children who were the most reactive during the venepuncture were those who had been identified by their parents as less sensitive and reactive to pain. These results are especially difficult to understand when considering that the notion of pain insensitivity in children with autism has been the product, in part, of studies that have used retrospective parental reports as the primary measure of pain sensitivity.^{1,5} Again, this may result from confusion on the part of the parents about pain in their children.

It might be argued that the behavioral measures used here (facial activity and behavioral distress) were not appropriate for this population and that greater weight should be given to parental reports over behavioral ratings. There are several difficulties with this argument. First, parents, in general, tend to underestimate pain in their children¹² and parents of children with cognitive impairment perceive decreased pain sensitivity in their children compared with nonimpaired children.²⁰ This raises concerns about the validity of using proxy reports of pain and the potential biasing effects of beliefs about pain insensitivity in these children. Second, facial activity is one of the most consistent and specific indicators of pain across all ages and levels of development.^{31,32,47} It seems unlikely that facial activity would be a valid indicator of pain in neonates, infants, children, adults, and the elderly but not in children with autism. The results of this study do provide validity for the use of facial activity as an indicator of pain in these children. In response to a noxious stimulus, children with autism displayed a significant increase in facial activity.

There is another possible explanation for the finding of discordance between retrospective parental reports and behavioral reactivity of the children with autism. When the parents were asked to complete the NCCPC and pain temperament item, they were asked to identify a particular painful incident their child had experienced that would provide a basis for their judgments. Many incidents used by parents focused on everyday types of pain (eg, falling down, bumping into things) rather than pain arising in medical situations. The presentation of pain in children with autism may differ depending on the setting (home vs. hospital) and the type of painful incident (everyday pain vs. medical procedures). While the results suggest that children with autism are not insensitive to pain, context-dependent pain indifference cannot 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. Children with autism are often resistant to changes in routine⁹ and, as a result, may react differently in a novel setting than they would in a familiar one. Severity of the autism might also play a role; children whose autism was more severe may be most indifferent in everyday situations but, on the other hand, more disturbed by the socially intrusive nature of the venepuncture procedure. In this study, parents were asked to consider intensity, but not the emotional (ie, anxiety) components, of their child's pain. There may have been an influence on parent ratings related to the anxiety component of pain or anxiety related to the clinical setting. Future research is needed to explore the concordance between parent report of pain sensitivity and observed reactivity of children with autism to every day painful incidents.

The results of the preliminary analysis comparing the behavioral reactions of children with autism and a group of nonimpaired children suggest that the 2 groups react to pain in a similar manner. There were no overall differences in the facial activity of the 2 groups of children over the entire procedure. The only significant difference was that the children with autism had greater facial activity during the needle segment than the children without autism. It is conceivable that the general communication and social deficits associated with the syndrome of autism resulted in a relatively greater automatic or spontaneous facial display. Deficiencies in higher cognitive mediation or less internal self-regulation could be associated with a failure to inhibit social display of facial activity and therefore greater pain facial activity.⁴⁸ This finding is similar to the outcome of a study finding that children with pervasive developmental delays exhibited more facial actions in response to aversive smells than nonimpaired children.⁴⁹ Children with pervasive developmental disorders were construed as not inhibiting their facial responses to noxious stimuli because they were not socialized to do so.⁴⁹ Alternatively, the increased facial activity could result from an inability of children with autism to regulate the expression of aversive emotions.⁶

Any conclusions resulting from comparing the reactions of children with autism to children without autism are made with caution due to the differences between procedures. The children with autism were bundled and received a venepuncture for the purposes of injection, whereas the comparison children were unbundled and received a venepuncture for the purposes of blood collection. These differences may limit the conclusions that can be drawn, but they do not invalidate the conclusion that the children with autism in this study reacted in a similar fashion as the children without autism. The pain stimulus for both groups was the same: a venepuncture. In many of the children with autism, the injection of the solution did not begin during the 10 seconds that were coded as the needle phase (the butterfly needle was in place, but the syringe

was not), so what was coded during the needle segment was the response to the venepuncture and not an injection. In addition, the children with autism and the comparison children did not differ in facial activity or behavioral distress during the baseline and pre-needle segments, suggesting that neither group was more distressed than the other. The significant main effect for behavioral distress for the children with autism was due to their being more distressed during the post-needle and recovery phases and not the bundling. Future research needs to be done using identical procedures to clarify these results.

Contrary to expectations, parents' retrospective accounts of their children's pain sensitivity and reactivity did not differ between the children with autism and the children without autism. An earlier study had reported that parents of children with cognitive impairment perceived decreased pain sensitivity in their children compared with nonimpaired children.²⁰ However, the parents of the children with autism in the current study were not asked to compare their children to children without autism. Rather, the parents were asked to characterize their children without reference to whatever they believed about other 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 of their children's pain and the objectively coded facial pain response was significantly greater for the comparison group than the autism group. In addition, for the comparison group, the correlations between retrospective parental judgments and observed behavioral reactions were positive but nonsignificant. This is in contrast to the autism group that (as reported earlier) had significant negative correlations between the observed behavioral responses of the children and the retrospective parental reports of pain sensitivity/reactivity. Further research is needed to explore the reasons for these differences.

The study had limitations comparable to many studies in the field of autism research. Opportunistic use of unobtrusive observation in clinical contexts resulted in an inability to control variables that may have influenced pain behaviors. For example, there were differences in the venepuncture procedures between the comparison and autism groups. While the pain stimulus (venepuncture) was similar between the groups, the contexts of the procedures differed. Although the comparison group used was not an ideal control, it is a beginning approximation given the almost complete lack of empirical research in this area. It is unlikely that a perfect control group could be found, given the clinical nature of this research. The difficulty of finding appropriate comparison groups remains a major challenge in understanding pain in children with autism, but this should not be a barrier to furthering our knowledge about pain in these children.

Finally, it needs to be emphasized that this 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.

CONCLUSIONS

The findings of this study indicate that children with autism should be viewed as being as reactive to painful stimuli as children without autism and do not support beliefs about pain insensitivity in children with autism. In response to a noxious stimulus, the children with autism in this study showed a substantial behavioral reaction. The difficulty in understanding pain in these children reflects the challenges adults encounter in decoding their pain behavior. Clinically, these results suggest that children with autism may be at risk for substandard pain management resulting from beliefs about pain insensitivity in these children. Additional research is needed to understand factors that may influence pain expression in this population and that may influence caregiver reports of pain.

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