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Commentary

Pain in children with Autism Spectrum Disorder: Experience, expression, and assessment

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Introduction

Historically, children with developmental disabilities have been excluded from pain research (Breau et al., 2006) despite comorbidities that increase risk of pain (Bottos & Chambers, 2006). Children with developmental disabilities and communication impairments may be particularly at risk as pain in individuals with communication impairments is frequently ignored and ineffectively managed (Hadjistavropoulos et al., 2001; Oberlander & Symons, 2006; Craig, 2009). One such population is children with Autism Spectrum Disorder (ASD). In the 5th edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5), ASD is conceptualized as a disorder involving difficulties in social communication and interaction as well as "restricted, repetitive patterns of behavior, interests, or activities"; this latter criterion includes "hyper- or hyporeactivity to sensory input or unusual interest in sensory aspects of the environment (e.g., apparent indifference to pain...)" (American Psychiatric Association, 2013).

Research on pain in children with ASD has generally depended on parent report of child pain (Bottos & Chambers, 2006); caregivers have frequently reported that children with cognitive impairment and/or ASD experience pain differently than typically developing children (Fanurik et al., 1999; Militerni et al., 2000; Inglese, 2008). In particular, there have been speculations that children with ASD display reduced sensitivity to pain (e.g. Cascio et al., 2008; Messmer et al., 2008; Tordjman et al., 2009)[1]. However, these contentions have occurred in the absence of definitive research findings. Arguing against pain insensitivity, Nader et al. (2004) found that during venipuncture, children with ASD displayed overall facial pain responses comparable to typically developing children and greater facial reactivity during the needle phase. The objectives of this commentary are to examine: (a) pain expression and experience in ASD, (b) challenges in pain assessment, (c) and the role of socio-communicative deficits and pain context.

Pain expression and experience

The way that a person expresses pain can give insight into his/her pain experience, but these are not identical constructs (Craig, 1986). Pain expression involves a person's observable response to a noxious stimulus, including pain behaviors, whereas a person's pain experience is internal and includes severity of discomfort (Craig, 1986). Children with ASD are often reported to respond differently to sensory stimuli compared to typically developing children; these responses are assumed to reflect differences in sensory experience and/or difficulties with sensory integration (Talay-Ongan & Wood, 2000; Watling et al., 2001; Baranek, 2002; Tomcheck & Dunn, 2007; Ashburner et al., 2008; Ben-Sasson et al., 2009; Klintwall et al., 2011). Unusual responses to sensory stimulation, including a potential indifference to pain, are described in the associated features and diagnostic conceptualizations of the disorder (McPartland et al., 2012; American Psychiatric Association, 2013).

Some researchers also suggest that children with ASD may express pain differently, but the direction and empirical bases of these differences are unclear (Militerni et al., 2000; Bottos & Chambers, 2006): studies report hypersensitivity (Nader et al., 2004; Tordjman et al., 2009), hyposensitivity (Militerni et al., 2000), or both hypo- and hypersensitivity (Inglese, 2008). Children with ASD may differ from typically developing children in general sensory sensitivity while having similar pain experiences (Minshew & Hobson, 2008). How do we understand these inconsistencies? Pain assessment method, socio-communicative deficits, and context may all play a role.

Pain assessment

Pain assessment is commonly achieved through self-report, observational, and/or proxy methods. While self-report is frequently used with typically developing children, impaired communication skills may make self-report questionable for populations with ASD (Breau & Burkitt, 2009; Craig, 2009). Recent studies (Minshew & Hobson, 2008; Bandstra et al., 2012) have gathered self-report from children and adolescents with ASD, supplemented with other measures (e.g. parent report, physiological measures). Findings indicate that some high-functioning individuals with ASD may be able to rate their own pain through self-report. However, ASD is a spectrum: just because high-functioning individuals with ASD are able to self-report their pain does not indicate that self-report is appropriate for the entire population.

Unfortunately, observational pain assessment can also be difficult due to idiosyncratic behaviors associated with ASD and/or cognitive impairment (e.g. atypical vocalizations, facial expressions) that may result in misestimates of pain by those unfamiliar with the individual's typical behavioral responses (Fanurik et al., 1999; Bottos & Chambers, 2006). Thus, different observers/caregivers may interpret pain behaviors differently (Coll et al., 2011; Fanurik et al., 1999). Additionally, there may be a disconnect between observed pain behavior and physiological response: Tordjman et al. (2009) found that while individuals with ASD were more likely to be rated as demonstrating an absence of pain reactivity by their behavior compared to a control group, individuals with ASD showed significantly greater heart rate before, during and after a venipuncture than the control group. An increase in heart rate is not necessarily specific to pain; anxiety or other forms of arousal may also cause a change in heart rate. Hence, while physiological measures such as heart rate do provide a helpful index of possible pain reactivity, they are also likely to be confounded by other types of physiological arousal and/or distress (e.g. anxiety).

Any proxy report of pain is likely to be based, at least partly, on observable behaviors (Coll et al., 2011). Much of the research on pain in children with ASD has been conducted using parent report due to communication challenges and difficulty self-reporting pain (Bottos & Chambers, 2006). However, parents of children with ASD may not be reliable in providing proxy reports of their children's pain, as their reports may be influenced by a variety of factors including information provided by health care professionals about the child's sensation and reaction to pain (Breau et al., 2003). Parents of children with cognitive impairment have been shown to believe that their children experience less pain than typically developing children (Breau et al., 2003); parents may hold similar beliefs regarding children with ASD. These beliefs may influence pain ratings: Dickie and colleagues (2009) found that parents of children with ASD were more likely than parents of typically developing children to attribute sensory responses as characteristic of ASD. Thus, parents of children with ASD may report their child's pain differently than parents of typically developing children but whether this reflects true differences in pain experience is unclear. In fact, parent reports have been shown to have no significant relationship with self-reported and psychophysically-measured sensory sensitivity in children with ASD (Güçlü et al., 2007). Similarly, during venipuncture, no relationship was found between parent ratings of child pain and facial responses of children with ASD, whereas there was a moderate relationship for typically developing children (Nader et al., 2004). Furthermore, strong negative correlations were found between parental report of their child's

general pain sensitivity/reactivity and observed behavioral responses of children with ASD; no such relationship existed for typically developing children (Nader et al., 2004). In sum, assessment method is important to consider when understanding the experience and expression of pain in this population.

Role of socio-communicative deficits and pain context

While it may be proposed that biophysical differences result in (suspected) differences in sensory thresholds and observable expression in populations with ASD, socio-communicative impairment may offer a logical explanation (Gilbert-MacLeod et al., 2000). Gilbert-MacLeod et al. (2000) found that developmentally delayed children were less likely to engage in help-seeking behavior following a painful event compared to non-delayed children, which may impair caregivers' ability to detect a child's pain. It would be beneficial for future studies to examine help-seeking behavior following painful episodes in children with ASD. Aberrant responses to the social environment, characteristic to individuals with ASD, may impact pain behavior and hinder caregivers' pain assessment and management (Nader et al., 2004; Inglese, 2008; Craig, 2009). Fitzgibbon and colleagues (2013) argue that in individuals with ASD, sensory pain processing abnormalities may be associated with impaired social processing, since both physical and social pain may be atypical in this population, and have overlapping neural correlates.

Numerous other factors may influence our understanding of pain in this population including cognitive functioning and presence of other psychological issues which could influence pain (Coll et al., 2011). There is considerable variability in the level of cognitive functioning of individuals with ASD and in research samples. For example, some research samples (e.g. Militerni et al., 2000; Nader et al., 2004; Tordjman et al., 2009) included low-moderate functioning individuals whereas Bandstra et al. (2012)'s sample was in the high functioning range. Research sample characteristics also have clear implications for the generalizability of the findings. Unfortunately, the current state of the literature does not allow conclusions about whether research findings on pain experience and expression reflect cognitive and developmental delays in the research samples rather than characteristics associated with ASD per se.

To understand inconsistencies in pain expressions, pain context may also be critical (Nader et al., 2004), particularly in populations with ASD given inflexibility to changes in routines and frequently comorbid anxiety (van Steensel et al., 2011; American Psychiatric Association, 2013). Pain behaviors in children with ASD may be more pronounced in some contexts than others (Tordjman et al., 2009). Sensory behaviors may be reported differently by the same caregiver in different contexts or over time: parents may be most reliable in reporting behaviors that are more frequent, intense or disruptive in terms of daily activities (Little et al., 2011).

Summary and suggestions for future research

In summary, it appears as though children with ASD may have difficulty integrating and interpreting sensory experiences, but it is unclear whether children with ASD actually express and/or experience pain differently than their peers. Our understanding of pain expression and experience are related to the characteristics of the individual as well as the assessment method. Children with ASD may not provide reliable verbal or behavioral cues for their experiences of pain, and parents of children with ASD may not be reliable in their proxy reports.

Unfortunately it can be difficult for others to meaningfully assess pain behaviors due to their own beliefs/biases, the child's idiosyncratic behavior and socio-communicative deficits, and assessment is likely affected by context. Pain assessment, which is foundational to understanding pain experience and guiding treatment, remains a challenge. Mistaken beliefs about pain sensitivity may lead to inadequate treatment of pain for children with ASD. Malviya et al. (2001) found that following surgery, pain assessment occurred less frequently for children with cognitive impairment, who also received less pain medication; while the authors speculated on a number of reasons for these findings including the potential of differential pain perception, it could have also been due to

clinicians' beliefs of altered pain perceptions of individuals with cognitive impairment. Future studies should examine these issues in a sample with ASD.

Much more systematic research with populations with ASD is needed before solid conclusions about the interrelationships of pain sensitivity, experience, expression, and behaviour can be made. Future research should:

- Examine pain sensitivity using multimodal assessment including brain imaging technologies such as functional magnetic resonance imaging (fMRI) or transcranial magnetic stimulation (TMS) (Fitzgibbon et al., 2013);
- Directly compare children with ASD with other populations;
- Examine the relationships among caregiver beliefs, their day-to-day experience with their child, and pain ratings for their own child vs. others with ASD;
- Control for cognitive impairment to understand its contributions to the issue while also considering to which subgroups of the ASD population the particular findings are generalizable.

It is recommended that clinicians use multimodal measures of pain (e.g. parent proxy, self-report, observational, physiological) when conducting pain assessments with this population. Furthermore, it is important that caregivers be particularly aware of painful events in individuals with ASD (Tordjman et al., 2009), since altered pain behavior/expression does not mean that there is altered pain sensitivity, experience or harm.

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Endnote

[1] Hyposensitivity, rather than hypersensitivity, has been emphasized in successive versions of the DSM, and also in the Sensory Experiences Questionnaire developed by Baranek et al. (2006) which contains an item asking about pain hyposensitivity only. The physiological mechanisms underlying pain and self-injurious behaviors in individuals with ASD and other developmental disabilities, such as differences in the release of endogenous opioids, are beyond the scope of this commentary (see Oswald et al., 1994, Canitano, 2006, Symons, 2011).

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