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Sensory modulation and negative affect in children at familial risk of ADHD

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A R T I C L E I N F O

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A B S T R A C T

Background/aims: Sensory modulation difficulties are commonly reported in patients with ADHD, however there has been little focus on the development of these difficulties in young children at a higher risk of later ADHD diagnosis. This study investigated whether children with a familial history of ADHD show greater sensory modulation difficulties. We also explored whether sensory modulation was linked to negative affectivity, which has been highlighted as a potential early marker of ADHD.

Methods: Parents of children under 6 years with a family history of ADHD (n = 65) and no family history (n = 122) completed questionnaires on sensory modulation and temperament.

Results: Children from families with ADHD were reported to display extreme patterns of hyperresponsiveness and hyporesponsiveness, relative to controls. No differences emerged for the sensory seeking domain. Some children within the high-risk group reported high scores across all three sensory modulation patterns. Regression analysis revealed that hyperresponsiveness predicted higher levels of negative affect.

Conclusions/implications: This study is the first to report greater sensory modulation difficulties in children at familial risk of ADHD. Future research should establish whether children with sensory modulation and temperament difficulties in early childhood are more vulnerable to developing ADHD.

What this paper adds?

This is the first study to investigate differences in patterns of sensory modulation in children with a family history of ADHD. Our results show that children at familial risk of ADHD experience more sensory modulation difficulties and more extreme patterns of hyperresponsiveness and hyporesponsiveness. Hyperresponsivity was also predictive of higher levels of negative affect. These findings highlight the need to consider sensory processing difficulties in the diagnosis and treatment of ADHD. Furthermore, this preliminary study demonstrates the need for further research to explore sensory modulation and temperament domains as potential early precursors to later ADHD symptomatology.

1. Introduction

Attention deficit hyperactivity disorder (ADHD) is a neurodevelopmental disorder characterised by persistent symptoms of inattention, hyperactivity, and impulsivity which impact on daily functioning (American Psychiatric Association, 2013). The estimated

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prevalence for ADHD in children worldwide is 5.9–7.1% (Polanczyk, Willcutt, Salum, Kieling, & Rohde, 2014). ADHD is a highly heritable disorder. One study found that up to 25%–30% of children with ADHD will have at least one parent affected by ADHD, and a larger percentage will have a parent with ADHD traits that do not meet full diagnostic criteria (Thissen et al., 2014). Another study reports a rate of 57 % chance of children developing ADHD in a sample of parents with childhood onset ADHD (Biederman et al., 1995). A three to five-fold increase in risk has been observed for children with siblings who have ADHD (Biederman et al., 1992; Faraone et al., 1993).

Sensory processing difficulties and emotional reactivity issues have frequently been reported in children with ADHD (Auerbach, Atzaba-Poria, Berger, & Landau, 2004; DeSerisy, Hirsch, & Roy, 2019; Dunn & Bennett, 2002). Children with ADHD can display both hyperresponsive and hyporesponsive behaviours in response to sensory stimuli, as well as difficulties with negative affect (Auerbach et al., 2008). However, it is unclear as to whether these co-occurring difficulties are present at an earlier stage of development in children at risk of ADHD. Further, little is known about how sensory modulation patterns are associated with temperament in this particular population.

1.1. *ADHD and sensory modulation*

In the 1970s, Ayres developed a theory of sensory integration to describe how the nervous system translates sensory information into responses (Lane et al., 2019). Ayres used the term sensory integration dysfunction to describe the disorder whereby impaired sensory processing can result in a variety of functional difficulties (Miller, Anzalone, Lane, Cermak, & Osten, 2007). Different models of sensory processing have developed since Ayres' original work such as Dunn (1997), (2014) and Baranek, Reinhartsen, and Wannamaker (2001). Miller and colleagues (2007) describe sensory integration dysfunction as a diagnostic disorder known as sensory processing disorder (SPD) and which can be broken down into three main patterns: sensory modulation disorder, sensory-based motor disorder, and sensory discrimination disorder. The most recent model of sensory processing comes from Bundy and Lane (2020) in which sensory processing dysfunction can be broken down into sensory modulation disorder and dyspraxia. The diagnosis of sensory processing disorder is included in the 0−5: Diagnostic Classification of Mental Health and Developmental Disorders in Infancy and Childhood (Zero to Three, 2016) and the Diagnostic Manual for Infancy and Early Childhood (Interdisciplinary Council on Development & Learning Disorders, 2005). However, it has not been classified in the Diagnostic Statistical Manual of Mental Disorders (DSM) (American Psychiatric Association, 2013) or the International Classification of Diseases (ICD) (World Health Organisation, 2018).

Recent research has seen a focus on sensory modulation since the introduction of hyper-and hyporesponsive behaviours as diagnostic criteria for autism spectrum disorder (ASD) in the DSM-V (American Psychiatric Association, 2013; Tavassoli et al., 2019). Sensory modulation concerns the ability to regulate and organize the degree, intensity, and nature of responses to sensory experiences in an appropriate manner (Lane, Miller, & Hanft, 2000). Sensory modulation occurs across all seven sensory systems: visual, auditory, tactile, olfactory, gustatory, vestibular, and proprioceptive. Many studies have focused on auditory and/or tactile processing systems alone (Parush, Sohmer, Steinberg, & Kaitz, 2007; Royeen & Fortune, 1990; Ghanizadeh, 2009). Nonetheless, it is important to explore all sensory domains to determine which domains are more likely to present difficulties in terms of planning effective treatment and intervention. Sensory modulation disorder (SMD) can present itself in three distinct but overlapping patterns: 1) hyperresponsivity or overresponsivity, 2) hyporesponsivity or underresponsivity, and 3) sensory seeking (Miller et al., 2007). Sensory hyperresponsivity relates to extreme, adverse or avoidant responses to sensory input (Tavassoli et al., 2019), similar to sensory sensitivity in Dunn (1997) model (Soto, Ciaramitaro, & Carter, 2018). Sensory hyporesponsivity is characterised by slow and muted responses to sensory stimuli (Mulligan et al., 2019). Sensory seeking concerns the craving of increased or more intense sensory input (Mulligan et al., 2019). Children can experience varying levels of all three patterns (Brock et al., 2012). The distinct pattern of sensory seeking has been questioned, with the suggestion that sensory seeking behaviours may be a compensatory mechanism to moderate high arousal levels (Liss, Saulnier, Fein, & Kinsbourne, 2006). For example, in Dunn (1997) model of sensory processing, the sensory seeking pattern acts as a modulator for a high sensory threshold to increase sensory stimulation.

Research has shown that children with ADHD are at a greater risk of experiencing sensory modulation problems (Ben-Sasson, Soto, Heberle, Carter, & Briggs-Gowan, 2017; Engel-Yeger & Ziv-On, 2011; Ghanizadeh, 2011; Mimouni-Bloch et al., 2018). Previous studies report that up to 65 % of children with ADHD also showed atypical sensory processing (Pfeiffer et al., 2015; Mimouni-Bloch et al., 2018). In one study of young children with ADHD, sensory processing difficulties were a significant predictor of ADHD (Yochman et al., 2006). It is important to note that there can be some overlap between symptoms of sensory hyper- and hyporesponsivity, and the core hyperactive behaviours in children with ADHD (Ben-Sasson et al., 2017; Panagiotidi, Overton, & Stafford, 2018). For example, the sensory seeking and hyperactive-impulsive (ADHD) subtypes can both be described by poor impulse control, and inappropriate movement and touch. The sensory overresponsivity and inattentive (ADHD) subtypes are both characterised by distractibility and difficulty focusing. Finally, the sensory underresponsivity and inattentive (ADHD) subtypes both include being unaware when spoken to or when asked to follow directions (Miller et al., 2012). However, studies comparing children with a dual diagnosis of sensory modulation difficulties and ADHD with children with only one of these disorders have shown that sensory modulation difficulties and ADHD behaviours can be reliably distinguished from each other based on both parent-reports and behavioural measures (Ben-Sasson et al., 2017; Reynolds, Lane, & Gennings, 2010; Yochman, Alon-Beery, Sribman, & Parush, 2013).

In children with ADHD, sensory modulation difficulties have been found to affect play, social participation, and increased functional impairment (Cosbey, Johnston, & Dunn, 2010; DeSerisy et al., 2019; Mangeot et al., 2001). Sensory modulation difficulties in children with ADHD can be related to increased delinquent and aggressive behaviours (Ben-Sasson et al., 2017; Mangeot et al., 2001). Early detection and management of sensory difficulties can help to improve functioning in children with ADHD (Ghanizadeh, 2011). As such, it is particularly important to attempt to identify and treat sensory modulation difficulties in this population (Wilkes-Gillan, Bundy, Cordier, Lincoln, & Chen, 2016). Given that these sensory difficulties can impact abilities in behavioural, emotional, motor, and cognitive domains, it is important to identify potential difficulties at an early stage (Jorquera-Cabrera, Romero-Ayusso, Rodgriguez-Gil, & Trivino-Suaraez, 2017). Previous research has shown that children who had both ADHD symptoms and sensory overresponsivity could be separated from typically developing children by higher sensitivity scores at 18 months (Ben-Sasson et al., 2017). This study was conducted with a community sample, used a dimensional approach, and focused only on auditory and tactile processing. Additional studies investigating sensory modulation across all sensory domains, and using both dimensional and diagnostic approaches, can provide greater clarity on the development and aetiology of the disorder (Dallos et al., 2017). Further evidence suggests that children with ADHD show sensory sensitivity in infancy before attention, impulsivity, or hyperactive symptoms emerge (Kaplan, Sadock, & Grebb, 1994). Thus, exploring sensory modulation in children at-risk of ADHD may inform whether early sensitivity is a risk marker for ADHD symptoms at school-age.

1.2. ADHD and negative affect

Putnam and colleagues have identified three factors of infant temperament: 1) surgency, 2) negative affect, and 3) effortful control (Putnam, Gartstein, & Rothbart, 2006). Negative affect is characterized by responses such as fear, anxiety, discomfort, and anger or frustration (Allan, Lonigan, & Wilson, 2013). Recent research indicates that markers of ADHD symptoms could be observed in early development, through parent-reported child temperament (Frick, Bohlin, Hedqvist, & Brocki, 2019; Miller et al., 2020). In particular, it has been theorised that high negative affectivity in early development may disrupt the development of effortful control, and by extension, act as an early marker for ADHD (Sullivan et al., 2015).

A significant proportion of children with ADHD have been found to experience emotional challenges such as poor emotional self-regulation, reduced empathy, and increased emotional reactivity to negative environmental events (Okado, Mueller, & Nakamura, 2016). A study of 66 male infants at familial risk of ADHD showed more anger reactivity at 7 months compared to typically developing controls (Auerbach et al., 2004). Similarly, 6-month-old infants with a family history of ADHD reported higher levels of distress than infants without a family history of the disorder (Sullivan et al., 2015). High negative affect was associated with ADHD symptoms of inattention, hyperactivity, and impulsivity in 3−6-year-old children (Martel, Gremillion, Roberts, Zastrow, & Tackett, 2014). Martel and colleagues (2014) suggest that negative affect may be particularly relevant during early development when neural circuitry is rapidly growing. Similarly, Wichstrøm, Penelo, Rensvik Viddal, de la Osa, and Ezpeleta (2018) observed that ADHD symptoms were predicted by high negative affect in a Norwegian sample of children.

Nigg, Goldsmith, and Sachek (2004) report that externalizing behaviour problems including conduct disorder and aggression can occur in more than 50 % of children with ADHD. High negative affect is associated with oppositional defiant disorder (ODD) (Eisenberg, Chang, Ma, & Huang, 2009; Okado et al., 2016). A similarly high co-morbidity rate has also been observed between ADHD and psychiatric disorders, many of which are characterized by symptoms of negative mood such as depression and anxiety (Lonigan, Phillips, & Hooe, 2003). Examining early emotional reactivity in children with a family history of ADHD may be one route to further understanding the disorder, particularly more complex cases.

1.3. Sensory modulation and negative affect

Deficits in sensory modulation have been associated with difficult temperament (Case-Smith, Butcher, & Reed, 1998; DeSantis, Coster, Bigsby, & Lester, 2004). Infant studies have observed a relationship between sensory processing and negative affect (DeGangi, Craft, & Castellan, 1991; DeSantis et al., 2004). Most often, sensory hyperresponsivity has been positively correlated with aspects of negative affect such as fear and irritability (Keuler, Schmidt, Van, Lemery-Chalfant, & Goldsmith, 2011). One study reports that tactile, visual, and auditory defensiveness, all of which are aspects of hyperresponsivity, were significantly related to irritable temperament in preterm infants (Case-Smith et al., 1998). Infants described as being fussy, irritable, and having poor attention skills were found to also present with hyperresponsivity to tactile, visual, and vestibular stimuli (DeGangi et al., 1991). A twin study of young children observed moderate correlations between negative affect and fear with auditory and tactile hyperresponsivity (Keuler et al., 2011). A similar result was observed by Goldsmith, Van Hulle, Arneson, Schreiber, and Gernsbacher (2006), in which low to moderate positive correlations were found between sensory hyperresponsivity and various temperament scales such as sadness, anger, and soothability. Fear showed a relatively high correlation with hyperresponsivity (Goldsmith et al., 2006). Sensory hyperresponsivity has been observed as a risk factor for anxiety disorders, with hyperresponsive preschool children also displaying high levels of anxiety symptoms at school age (Carpenter, Baranek, & Copeland, 2019). Early sensory hyperresponsivity has also been found to predict later anxiety in autistic children (Green, Ben-Sasson, Soto, & Carter, 2012). Treatments for sensory responsivity has been successful in decreasing anxiety in adults and young autistic children (Baranek et al., 2015; Pfeiffer & Kinnealey, 2003). Given the high co-occurring rate of anxiety and ADHD, (Reynolds & Lane, 2009) investigating whether sensory responsivity is contributing to this relationship could inform treatment and intervention options for this population. In the current study, we expect that higher levels of negative affect will be associated with higher levels of sensory hyperresponsivity.

In contrast to the body of literature examining hyperresponsivity and temperament, little focus has been paid to the relation between both sensory seeking and sensory hyporesponsivity and negative affect. One study in an ASD population found an association between both hyporesponsivity and sensory seeking patterns and more negative mood (Brock et al., 2012). As such, within an at-risk ADHD population, it is unclear whether a relation exists between the hyporesponsivity and negative affect, and which direction this relationship might be. Similarly, it is unclear whether a relation exists between sensory seeking and negative affect in this population.

To our knowledge, no empirical studies have examined the relation between negative affect and sensory modulation in children at

Table 1
Demographic breakdown of sample.

		High-risk (n = 47) M (SD)	Low risk (n = 98) M (SD)	p value
Age		3.40(1.61)	2.42(1.56)	.001
		N (%)	N (%)	
Gender	Male	28(59.6 %)	67(68.4 %)	.300
	Female	19(40.4 %)	31(31.6 %)	
Ethnicity	Irish/British White	41(87.2 %)	90(92.8 %)	.403
	Mixed race	5(10.6 %)	8 (6.6 %)	
	Other	1(2.1 %)	2(2.1 %)	
Single Parent?	Yes	10(21.3 %)	4(4.1 %)	.009
Maternal Education	Junior Cert/GCSE	4(11.1 %)	2(2.2 %)	.003
	Leaving Cert/A Levels	5(13.9 %)	2(2.2 %)	
	Level 6 Cert	3(8.3 %)	6(6.6 %)	
	Undergraduate	11(30.6 %)	23(25.3 %)	
	Postgraduate	13(36.1 %)	58(63.7 %)	
Normal Pregnancy		37(78.7 %)	73(74.5 %)	.580
Post Natal Depression		11(23.4 %)	14(14.3 %)	.208
ASD in immediate family		18(38.3 %)	2(2.0 %)	.000

familial risk of ADHD. This study considers how sensory modulation is associated with early temperament markers of ADHD in children at familial risk for the disorder.

1.4. The current study

The current study aims to examine sensory modulation in children between the age of 7 and 72 months with a higher likelihood of developing ADHD due to familial diagnosis. This age range was chosen as it is old enough to capture the temperament component of negative affect (Putnam, Rothbart, & Gartstein, 2008) yet younger than the typical age range of ADHD diagnosis at 6–12 years of age (Miller et al., 2020; Visser et al., 2014). A secondary aim of this study is to explore the relation between sensory modulation, familial risk of ADHD, and the temperament dimension of negative affect. While previous studies have reported a link between poor sensory modulation and negative affect, no study to date has looked at this relationship in children at-risk of later ADHD diagnosis. Understanding this relation between negative affect and sensory modulation in children at-risk of ADHD is important to identify whether a certain population of children (i.e. those with sensory processing problems and negative emotional reactivity) could be at greater risk of developing later ADHD. Based on previous research, the following research questions were proposed:

Research question 1: Do parents of children at-risk of ADHD report poorer sensory modulation than children without a family history of ADHD. In line with previous research in older children with ADHD, it is expected that parents of children at-risk for ADHD will report more sensory modulation difficulties than parents of young children with no family history of ADHD.

Research question 2: Is there an association between sensory modulation, familial risk of ADHD, and negative affect? It is expected that children with greater levels of hyperresponsivity will also have high levels of negative affect. In terms of hyporesponsiveness and sensory seeking, it is unclear which direction the relationship will be in, if any relationship does exist.

2. Method

2.1. Participants

The study was advertised through social media channels and parenting websites, on flyers in public spaces, and at ADHD events (e.g. parent talks). Parents were eligible to take part if they had a child aged between 6 and 72 months who was born full-term, had no history of brain injury, or epilepsy. Brain-based disorders and medical complications relating to prematurity may have an influence on brain development and sensory processing (Chorna, Solomon, Slaughter, Stark, & Maitre, 2014). Participants with an ASD diagnosis in their immediate family (which may also influence sensory processing difficulties) were not excluded from analyses due to the high co-occurring rate between the two disorders (Jang et al., 2013). Family history of ASD was considered in further analyses to investigate the influence of this risk factor on sensory processing scores. Parents self-recruited to the study by following a link to the questionnaire on Qualtrics software.

Twenty one children were removed from analysis due to not meeting the inclusion criteria: 14 were born preterm (n = 4 high-risk, n = 10 low-risk); 2 due to an epilepsy diagnosis (n = 1 high-risk, n = 1 low-risk), 1 due to a brain injury (n = 1 low-risk), 6 were over the age of 6 (n = 5 high-risk, n = 1 low-risk), and 1 for being under 6 months (n = 1 low-risk). One hundred and forty-five caregivers were included in the final analyses. Parents were asked whether there was a confirmed diagnosis of ADHD in the family and to indicate which family member received this diagnosis. Children were then divided into a high-risk or low-risk group based on family history of ADHD. Those in the high-risk group (n = 47) had a history of ADHD in the immediate family (i.e. a parent (29.8 %), sibling (57.42 %) or both parent and sibling (12.8 %) had an existing diagnosis). The low-risk group (n = 98) had no immediate family history of ADHD. Children's ages ranged from 7 months to 6 years 0 months (M = 2.74 months, SD = 1.64). In the high-risk group, of the 18 families with co-occurring ASD and ADHD, 14 had a sibling diagnosis of both ADHD and ASD, three had parents with a dual diagnosis, and one case where both a parent and sibling had ASD and ADHD. See Table 1 below for a demographic breakdown of the sample.

2.2. Measures

Parents first completed a short questionnaire which asked for demographic information, including maternal level of education, marital status, pregnancy, postnatal depression, and family history of mental health and neurodevelopmental disorders.

2.2.1. Sensory processing

The Sensory Experiences Questionnaire (SEQ) Version 2.1 (Short Form) is a caregiver questionnaire examining sensory processing in children up to 6 years (Baranek, 2018). The questionnaire contains 33 quantitative items which focus on three patterns of sensory response: hyperresponsiveness, hyporesponsiveness, and sensory seeking. The SEQ also has five subscales relating to the sensory domains of auditory, tactile, visual, olfactory/gustatory, vestibular/proprioceptive. The SEQ has good psychometric properties, with an internal consistency score of $a = 0.80$ and test re-test reliability of ICC = 0.92 (Little et al., 2011). This questionnaire was developed for use with children with autism and related developmental disorders (Baranek, 2018). To our knowledge, this is the first use of this measure in a population at-risk of ADHD. Higher scores reflect greater difficulties in sensory processing. Individuals scoring between 1 and 2 standard deviation above the mean are considered to be "at risk" for sensory processing difficulties. Scores greater than 2 standard deviations above the mean are considered to be in the "deficient" range for sensory processing.

2.2.2. Temperament

The Infant Behaviour Questionnaire (Very Short Form) (IBQ), the Early Childhood Behaviour Questionnaire (Very Short Form) (ECBQ) and the Childhood Behaviour Questionnaire (Very Short Form) (CBQ) were used to measure temperament. The IBQ-VSF is a caregiver report measure designed for use with children from 3 to 12 months of age, the ECBQ-VSF is designed for use with children from 18 to 36 months of age and is also recommended for use for children between 13–17 months of age (Putnam & Rothbart, 2006). The CBQ-VSF is the older version of the form designed for 3–8-year-old children (Putnam & Rothbart, 2006). All three measures are formed of three subscales of surgency, effortful control, and negative affect. The reliability of negative affect subscales across the measures ranged from .70−.78. Scores on the negative affect subscale from the IBQ, ECBQ and CBQ were transformed into Z-scores in order to compare across groups. Negative affect has been shown to be generally stable across development (Carranza, González-Salinas, & Ato, 2013; Putnam et al., 2008). The negative affect subscale contains questions such as how a child reacts to unfamiliar persons, their frustrations, and soothability.

2.3. Procedure

Ethical approval for the study was obtained from University College Dublin Human Research Ethics Committee. All parents provided informed consent before beginning the experiment. Questionnaires were completed online using Qualtrics software (Qualtrics, 2005). Data collection commenced in February 2018 and was completed in March 2019. Participants first provided demographic information before completing the IBQ, ECBQ or CBQ (depending on the age of the child) and the SEQ. Some parents did not complete all questionnaires (81.3 % of participants completed the SEQ only). Those who did not complete the temperament measure did not differ from those who did on maternal education, marital status, or family size. This study formed part of a larger study investigating a range of potential early markers for ADHD in children at familial risk of diagnosis.

2.4. Statistical analysis

All statistical analyses were conducted using SPSS 24 (SPSS Inc.). To address the first research question, independent t-tests were carried out between the high-risk and low-risk groups to investigate differences in sensory processing patterns and sensory processing domains. Effect sizes for significant differences were reported with Cohen's d, where 0.2 = small effect, 0.5 = medium effect, 0.8 = large effect (Cohen, 1988). An independent *t*-test was also carried out between the high-risk and low-risk groups for the negative affect subscale of the temperament measure. To address the second research question, correlations were carried out between sensory processing patterns and negative affect. A hierarchical regression analysis was then conducted to determine whether sensory processing patterns and group membership predict negative affect. For the regression model, there was independence of residuals, as measured by a Durbin-Watson statistic of 2.021. Homoscedasticity was present, as assessed by visual inspection of a plot of studentized residuals versus unstandardized predicted values. There was no evidence of multicollinearity, with all tolerance values greater than 0.1, with no correlations higher than 0.8 (Berry, Feldman, & Stanley Feldman, 1985; Neter, Kutner, Nachtsheim, & Wasserman, 1996). There were no studentized deleted residuals greater than ±3 standard deviations, no leverage values greater than 0.2, and values for Cook's distance above 1. The assumption of normality was met, as assessed by a Q-Q Plot. Variables were added in three stages. Age as a continuous variable was added first to control for age-related differences. The second variable added to the model was group membership, as we expect that children with a diagnosis of ADHD in their family will show higher levels of negative affect in each respective model. Hyperresponsivity and hyporesponsivity scores from the SEQ were then added to the regression in the final stage.

3. Results

3.1. Reliability analysis

Reliability analysis was carried out on all scales. The IBQ, ECBQ, and CBQ had moderate reliability, with Cronbach's alpha of .687,

Table 2
Independent *t*-test of SEQ subscales and negative affect between high-risk and low-risk groups.

Variable	High-risk M (SD) N = 47	Low-risk M (SD) N = 98	*t*	*p*	*d*
SEQ Total	70.19 (23.49)	67.02 (16.76)	.830	.410	.16
SEQ Hyporesponsiveness	11.64 (5.08)	9.88 (3.73)	2.116	.038*	.39
SEQ Hyperresponsiveness	27.32 (9.15)	24.20 (6.68)	2.082	.041*	.39
SEQ Sensory Seeking	31.23 (12.52)	33.01 (11.06)	−.865	.389	.15
SEQ Tactile	20.45 (7.14)	18.88 (5.92)	1.307	.195	.24
SEQ Auditory	14.96 (4.50)	13.76 (3.71)	1.702	.091	.29
SEQ Visual	11.36 (4.74)	11.62 (3.77)	−.330	.742	.06
SEQ Olfactory/Taste	11.72 (4.84)	11.36 (3.60)	.462	.646	.08
SEQ Vestibular/Proprioceptive	9.81 (4.01)	9.79 (3.60)	.034	.973	.01
Negative affect	.21(.98)	−.13(.90)	1.819	.072	.36

* Significant result ($p < .05$).

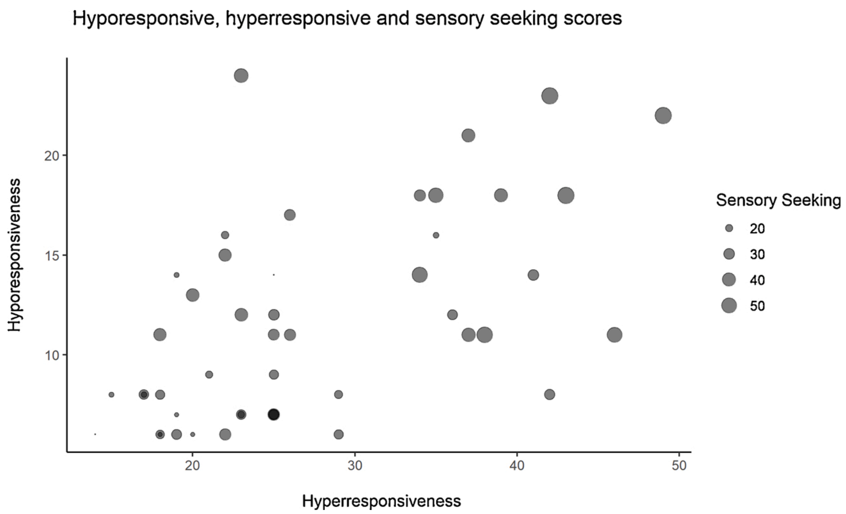

Fig. 1. Bubble plot of hyporesponsive, hyperresponsive, and sensory seeking scores for high-risk group .

.690, and .633 respectively. The SEQ had the strongest internal reliability, with Cronbach's alpha of .886.

3.2. *Sensory processing*

An independent *t*-test was carried out between the high-risk and low-risk groups to determine whether the groups were significantly different in relation to sensory processing patterns (Table 2). Results showed a significant difference between the groups in terms of SEQ hyporesponsiveness ($p = .038$, $d = 0.39$) and SEQ hyperresponsiveness ($p = .041$, $d = 0.39$). No difference was observed for the sensory seeking pattern or SEQ total score. As there were differences observed for two of the three sensory patterns, specific domains were explored, and no significant differences emerged. A significant portion of both groups scored greater than 1 standard deviation from the norm for overall sensory processing score (high-risk: 34.0 %; low-risk: 27.5 %). For hyporesponsiveness, the high-risk group had significantly more children scoring outside the normal range 55.2 %) compared to the low-risk group (29.6 %). For hyperresponsivess, 31.9 % scored outside the normal range compared to 16.3 % of low-risk children, however this difference was not significant. More children from the low-risk group than the high-risk group scored in the 'at-risk' or 'deficit' categories for the sensory seeking pattern, indicating scores greater than 1 standard deviation from the mean (33.6 % and 27.5 % respectively), although this difference was not statistically significant. In all sensory patterns except sensory seeking, children from the high-risk group reported higher scores, indicating that some children at-risk of ADHD experience more extreme patterns of sensory processing.

Correlations were also carried out for the subscales of the SEQ (see Fig. 1), as previous research has suggested that children can show varying levels of all three patterns (Brock et al., 2012). Results showed significant correlations between all three subscales: hyperresponsiveness and hyporesponsiveness ($r = .581$, $p < .001$); hyperresponsiveness and sensory seeking ($r = .474$, $p < .001$); and hyporesponsivess and sensory seeking ($r = .356$, $p < .001$). 21.3 % of children from the high-risk group are showing high levels of hyperresponsivity, hyporesponsivity, and sensory seeking. In contrast, only 3.1 % of the low-risk group had elevated scores across all three patterns (Fig. 2).

Within group differences for the high-risk group were also examined, based on family history of ASD. There were no significant differences between these groups on any of the subscales of the SEQ, the total SEQ score, or sensory domain scores.

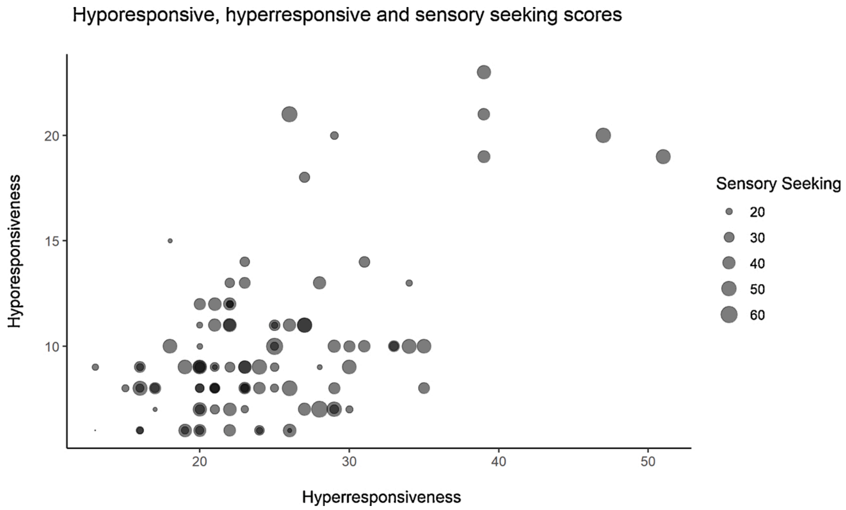

Fig. 2. Bubble plot of hyporesponsive, hyperresponsive, and sensory seeking scores for low-risk group .

Table 3
Hierarchical regression of age, group membership, and sensory processing predicting negative affect.

Negative Affect			
	Step 1	Step 2	Step 3
R^2	.024	.052	.185
F	2.721	2.960	6.008**
Age	.156	.117	−.032
Group	—	.171	.141
SEQ Hypo	—	—	−.110
SEQ Hyper	—	—	.455**

Note: coefficients shown are standardized coefficients. *p < .05, **p<.01.

3.3. *Negative affect*

No significant difference emerged between the high-risk and low-risk groups for negative affect levels, ($p = .072$;see Table 2).

3.4. *Associations between sensory processing patterns and negative affect*

For the whole sample, significant correlations were observed between negative affect and the overall SEQ score ($r = .316$, $p = .001$), the hyperresponsiveness subscale ($r = .382$, $p < .001$), and the sensory seeking subscale ($r = .201$, $p = .031$). There was a trend towards significance for the hyporesponsiveness pattern ($r = .181$, $p = .052$).

For the high-risk group, a significant correlation was observed between negative affect and total SEQ score ($r = .545$, $p < .001$), the hyperresponsiveness subscale ($r = .593$, $p < .001$), the hyporesponsiveness subscale ($= .364$, $p = .027$), and the sensory seeking subscale, ($r = .432$, $p = .008$). In contrast, no significant correlations emerged between negative affect and sensory scores for the low-risk group. A trend towards significance was observed between negative affect and the hyperresponsiveness subscale for the low-risk group ($r = .214$, $p = .059$).

A hierarchical regression was carried out to determine whether sensory processing patterns and group membership could predict negative affect, when controlling for age across the whole sample (Table 3 below). Age was controlled for as the two groups were not matched for age. The addition of group membership did not lead to a statistically significant change in R^2. The full model of age, group membership, SEQ hyperresponsivity, SEQ hyporesponsivity was statistically significant, with an increase in R^2 of 0.127, $F(4, 106) = 6.008$, $p < .001$, $R^2 = .185$, adjusted $R^2 = .154$. Taken together, all of the variables accounted for 18.5 % of variance in negative affect. Hyperresponsiveness was the only significant contributor to the model ($\beta = .455$, $p = .000$). Higher levels of hyperresponsivess were associated with higher negative affect.

4. Discussion

This study examined differences in sensory modulation between children with and without a familial history of ADHD. Findings showed that children at high-risk of ADHD were more likely to show both a hyperresponsive and hyporesponsive pattern of sensory processing, relative to children at low-risk of ADHD. This study is the first to report a higher rate of sensory modulation difficulties, which have already been established to be salient for children with ADHD, in young children at-risk of a later ADHD diagnosis due to familial history. A significant difference in sensory hyperresponsiveness between high-risk and low-risk children is concurrent with the literature on children with a confirmed diagnosis of ADHD. In the current study, 31.9 % of children at-risk of ADHD reported at-risk/deficient scores of hyperresponsiveness. Reynolds et al. (2010) observed that 13 of their 24 participants (54.17 %) with ADHD also had sensory hyperresponsivity. Similarly, Ben-Sasson et al. (2017) report that 48 % of a community sample who met the criteria for an ADHD diagnosis also met the criteria for hyperresponsivity. One study in an adult ADHD population report that 19 % of participants were hyporesponsive, while further 9.5 % of participants met the criteria for both hyporesponsiveness and hyperresponsivess (Bijlenga, Tjon-Ka-Jie, Schuijers, & Kooij, 2017). In the current study, a large portion of high-risk children (55.2 %) reported extreme patterns of hyporesponsiveness (falling into the at-risk or deficient range), and 29.8 % of children at-risk for ADHD showed extreme levels of both hyporesponsiveness and hyperresponsiveness. In comparison, only 7.1 % of children from the low-risk group displayed extreme levels of both hyporesponsiveness and hyperresponsiveness, while 29.6 % displayed at risk/deficient scores for hyporesponsiveness only. This is a large proportion of children (almost 30 %) showing at-risk/deficient scores for hyporesponsiveness and it may be that the SEQ overestimates risk. Parents who suspected sensory processing difficulties in their children may also have been more motivated to take part in the study, and this may explain the elevated hyporesponsiveness scores.

Children at-risk for ADHD did not show significantly more sensory seeking behaviours, which might be expected for this population. Previous studies have shown increased incidence of the sensory seeking pattern in children with ADHD (Engel-Yeger & Ziv-On, 2011; Ermer & Dunn, 1998; Mimouni-Bloch et al., 2018). However, typically developing young children often demonstrate increased sensory seeking behaviours, and these generally reduce by 4 years of age (Leekam et al., 2011). As the low-risk sample in the current study is significantly younger than the high-risk group, this might account for the absence of a significant difference between groups on the sensory seeking domain.

An association was observed between hyperresponsivity and negative affect, similar to findings from previous research (DeGangi et al., 1991; Goldsmith et al., 2006). Child's age and familial risk for ADHD was not related to negative affect, while hyperresponsivity accounted for 18.5 % of variance in negative affect levels. According to Cohen (1988), these adjusted R^2 values indicate a small to moderate effect size. Results indicate that higher levels of hyperresponsiveness predict higher levels of negative affect. This differs from a similar study examining the relation between sensory processing and temperament in a population with autism spectrum disorder (ASD). Brock and colleagues (2012) reported significant effects of association between a combination of sensory features (across all three patterns) and increased negative mood. A significant association was observed between hyporesponsivity and slowness to adapt, low reactivity, and low distractibility. No significant main effects were found between hyperresponsiveness or sensory seeking and the temperament subscales (Brock et al., 2012).

Similar to previous research in infants at-risk of ADHD (Auerbach et al., 2004), children with familial risk of ADHD showed higher levels of negative affect than children with a low-risk of ADHD diagnosis. In the present study this was a trend towards a significant difference (p=.072), which may be due to our small sample size. It has been suggested that the high levels of negative affect and negative emotionality observed in ADHD populations may be due to comorbid antisocial tendencies (Martel & Nigg, 2006). However, the presence of high levels of negative affect observed in this study at a young developmental age – taken with the results of the infant study by Auerbach and colleagues (2004) – suggest that high levels of negative affect may be related to the core symptoms of ADHD. This supports the dual-pathway temperament model of ADHD proposed by Nigg et al. (2004).

4.1. Limitations

Certain limitations need to be taken into account when considering the current findings. The SEQ was developed for an autism population and related developmental disorders (Baranek, 2018). As a result, this measure has not been validated prior to this study for use with a population at-risk of ADHD. Future research should further investigate the valid use of the SEQ in an ADHD population, and its psychometric properties. Furthermore, a significant portion of the high-risk group (38.3 %) also had a family history of ASD. This could be a confounding factor as hyporesponsiveness and hyperresponsiveness have been well established in the ASD phenotype (APA, 2013). The results of this study found no difference between those with a family history of both ASD and ADHD and those with ADHD only, suggesting that sensory modulation difficulties are salient for children at-risk of ADHD. Further research using larger samples of children with a history of ADHD only, those with a history of ADHD and ASD, and children with no family history of either disorder are required to further elucidate this overlap. Sensory modulation difficulties may explain part of the shared phenotype between ADHD and ASD (Dellapiazza et al., 2020). Given that sensory modulation difficulties are also present across a range of neurodevelopmental disorders (e.g. Developmental Coordination Disorder; Delgado-Lobete, Pértega-Díaz, Santos-del-Riego, & Montes-Montes, 2020) further research into early sensory processing may form part of the explanation for the underlying etiology of atypical development.

The majority of the sample is Irish/British white and well educated with an undergraduate or postgraduate qualification. This limits the generalisability of the findings to a wider population. It should also be noted that the groups were not matched for parental level of education, marital status, or age. Although, the two groups were matched in terms of gender and ethnicity, with age being controlled for in regression analyses. Furthermore, as the age range of the sample included participants who completed the IBQ, ECBQ and the CBQ (based on age cut-off ranges), it was necessary to calculate Z-scores in order to analyse temperament scores. The IBQ, ECBQ, and CBQ have been found to show stability across measures, in particular the construct of negative affect (Putnam et al., 2006). Although, it should be noted that the Cronbach's alpha for each scale in the current study was low (ranging from .63 to .69). As such, an element of caution is advised in interpreting the results of this study and future studies should aim to replicate these results with alternative measures of negative affect. An additional limitation is that the measures in this study are proxy-report. As such, they are open to issues such as questions being misunderstood, inaccurate recall, or response bias (Gorber, Tremblay, Moher, & Gorber, 2007). On the other hand, caregiver reports have a low participant burden and may be able to highlight behaviours not easily observable in other testing measures (Ermer & Dunn, 1998; Yochman, Parush, & Ornoy, 2004).

A recent study reported moderate associations between sensory modulation difficulties and ADHD symptoms in children with sensory modulation disorder and their parents (Kalig-Amir, Berger, Rigbi, & Bar-Shalita, 2019). Previous studies have also observed a relation between sensory modulation and ADHD in adults (Bijlenga et al., 2017; Panagiotidi et al., 2018). It is possible that children in the current study show difficulties in sensory modulation due to the fact that their parents have a high prevalence of sensory modulation difficulties in addition to ADHD. Perhaps future research should include a self-report measure of sensory processing to attempt to measure parent's own sensory modulation issues.

4.2. Future research and implications

Due to the current findings which show that sensory modulation difficulties can be present in children at familial risk of ADHD, sensory functioning should be considered in the evaluation of children who are at risk of ADHD (DeSerisy et al., 2019). Although, it is important to note that not all of the children in the high-risk group will later receive an ADHD diagnosis. However, it seems that a significant portion of children with ADHD do present with sensory modulation difficulties, and this subgroup may require a certain intervention approach (Ghanizadeh, 2011). Early detection and treatment of these difficulties can help children with ADHD in terms of their participation in social activities or prevent the development of secondary social-emotional problems, such as anxiety (Reynolds & Lane, 2009; Yochman et al., 2004). Previously, sensory interventions have been successful in reducing anxiety (an element of negative affect) in typical adults and autistic children (Baranek et al., 2015; Pfeiffer & Kinnealey, 2003). If early sensory modulation difficulties could be treated through targeted interventions, this might reduce negative affect levels in children and in turn, reduce later ADHD symptomatology. Understanding early sensory processing and the aetiology of sensory modulation is an important avenue for future research and early intervention planning.

Further research is required to determine how many children at familial risk for ADHD will receive a confirmed diagnosis of ADHD. Sensory processing abilities should also be assessed at this follow-up stage to discover if these group differences still exist at a clinical level. Similarly, additional longitudinal studies are needed to examine how negative affect can lead to later elevated ADHD symptomatology (Sullivan et al., 2015). It could be possible to establish whether children with poor sensory modulation abilities and difficult temperament in early childhood are more vulnerable to developing ADHD, given a family history of ADHD. Thus, it may be pertinent to prioritise this group in terms of intervention and treatment.

4.3. Conclusion

Previous research has shown that children with an ADHD diagnosis are likely to experience extreme patterns of sensory processing that can interfere with daily life and exacerbate the functional impairments of ADHD (Ben-Sasson et al., 2017; Mimouni-Bloch et al., 2018). To the author's knowledge, this study is the first to demonstrate that these extreme patterns of hyperresponsivity and hyporesponsivity are present in children at familial risk of ADHD. This study also found that higher levels of hyperresponsivity were predictive of higher levels of negative affect in this cohort. Recent research has identified the temperament domain of negative affect as a potential early marker to later ADHD symptomatology (Auerbach et al., 2008; Sullivan et al., 2015). As a result, it is important to consider whether young children showing high levels of hyperresponsivity may be particularly vulnerable to later ADHD diagnosis. This is an important potential avenue for early intervention, as treating early sensory modulation difficulties could reduce negative affect levels and severity of later ADHD symptomatology. Further research in this population is required to establish whether sensory modulation difficulties are a precursor to ADHD symptomatology.

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CRediT authorship contribution statement

J. Keating: Conceptualization, Methodology, Investigation, Formal analysis, Writing - original draft. **J. Bramham:** Conceptualization, Methodology, Supervision, Writing - review & editing. **M. Downes:** Conceptualization, Methodology, Supervision, Writing - review & editing, Funding acquisition.

Declaration of Competing Interest

The authors report no declarations of interest.

Appendix A. Scatterplots of SEQ subscale scores

Figs. A1–A3

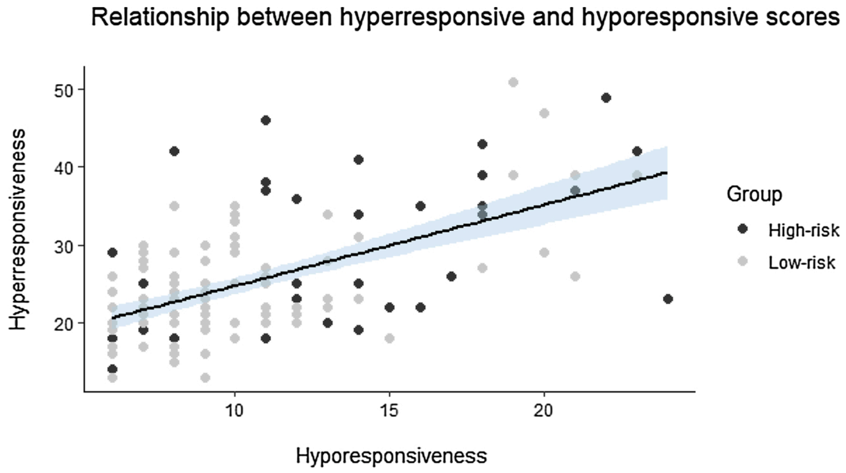

Fig. A1. Scatterplot of relationship between hyperresponsive and hyporesponsive scores on the SEQ.

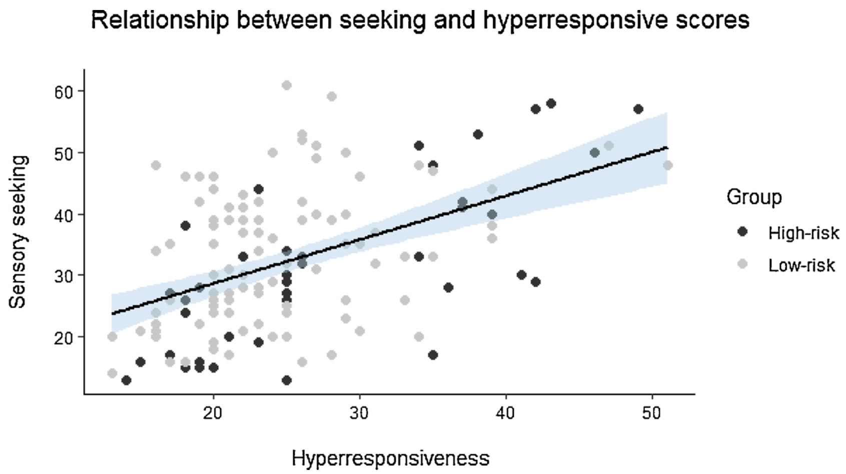

Fig. A2. Scatterplot of relationship between hyperresponsive and sensory seeking scores on the SEQ.

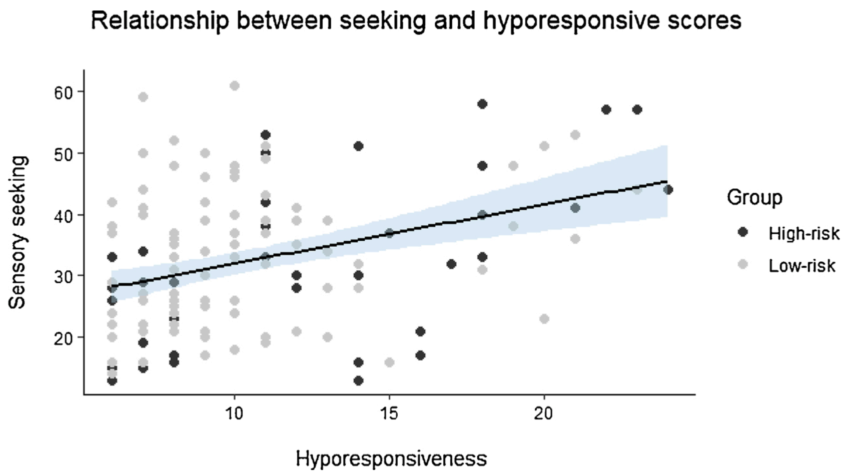

Fig. A3. Scatterplot of relationship between hyporesponsive and sensory seeking scores on the SEQ.

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