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23 **What this paper adds?**

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

32

33 **1. Introduction**

34 Attention deficit hyperactivity disorder (ADHD) is a neurodevelopmental disorder
35 characterised by persistent symptoms of inattention, hyperactivity, and impulsivity which
36 impact on daily functioning (American Psychiatric Association [APA], 2013). The estimated
37 prevalence for ADHD in children worldwide is 5.9 to 7.1% (Polanczyk et al., 2014). ADHD
38 is a highly heritable disorder. One study found that up to 25% to 30% of children with ADHD
39 will have at least one parent affected by ADHD, and a larger percentage will have a parent
40 with ADHD traits that do not meet full diagnostic criteria (Thissen et al., 2014). Another
41 study reports a rate of 57% chance of children developing ADHD in a sample of parents with
42 childhood onset ADHD (Biederman et al., 1995). A three to five-fold increase in risk has
43 been observed for children with siblings who have ADHD (Biederman et al., 1992; Faraone
44 et al., 1993).

45 Sensory processing difficulties and emotional reactivity issues have frequently been reported
46 in children with ADHD (Dunn & Bennett, 2002; Auerbach et al., 2004; DeSerisy et al.,
47 2019). Children with ADHD can display both hyperresponsive and hyporesponsive
48 behaviours in response to sensory stimuli, as well as difficulties with negative affect
49 (Auerbach et al., 2008). However, it is unclear as to whether these co-occurring difficulties
50 are present at an earlier stage of development in children at risk of ADHD. Further, little is
51 known about how sensory modulation patterns are associated with temperament in this
52 particular population.

53 **1.1 ADHD and sensory modulation**

54 In the 1970s, Ayres developed a theory of sensory integration to describe how the nervous
55 system translates sensory information into responses (Lane et al., 2019). Ayres used the term
56 sensory integration dysfunction to describe the disorder whereby impaired sensory processing
57 can result in a variety of functional difficulties (Miller et al., 2007). Different models of
58 sensory processing have developed since Ayres' original work such as Dunn (1997, 2014)
59 and Baranek et al., (2001). Miller and colleagues (2007) describe sensory integration
60 dysfunction as a diagnostic disorder known as sensory processing disorder (SPD) and which
61 can be broken down into three main patterns: sensory modulation disorder, sensory-based
62 motor disorder, and sensory discrimination disorder. The most recent model of sensory
63 processing comes from Bundy and Lane (2020) in which sensory processing dysfunction can

64 be broken down into sensory modulation disorder and dyspraxia. The diagnosis of sensory
65 processing disorder is included in the 0-5: Diagnostic Classification of Mental Health and
66 Developmental Disorders in Infancy and Childhood (Zero to Three, 2016) and the Diagnostic
67 Manual for Infancy and Early Childhood (Interdisciplinary Council on Developmental and
68 Learning Disorders, 2005). However, it has not been classified in the Diagnostic Statistical
69 Manual of Mental Disorders (DSM) (APA, 2013) or the International Classification of
70 Diseases (ICD) (World Health Organisation, 2018).

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

93

94 Research has shown that children with ADHD are at a greater risk of experiencing sensory
95 modulation problems (Ghanizadeh, 2011; Ben-Sasson et al., 2017; Mimouni-Bloch et al.,
96 2018; Engel-Yeger & Ziv-On, 2011). Previous studies report that up to 65% of children with
97 ADHD also showed atypical sensory processing (Pfeiffer et al., 2015; Mimouni-Bloch et al.,
98 2018). In one study of young children with ADHD, sensory processing difficulties were a
99 significant predictor of ADHD (Yochman et al., 2006). It is important to note that there can
100 be some overlap between symptoms of sensory hyper- and hyporesponsivity, and the core
101 hyperactive behaviours in children with ADHD (Ben-Sasson et al., 2017; Panagiotidi et al.,
102 2018). For example, the sensory seeking and hyperactive-impulsive (ADHD) subtypes can
103 both be described by poor impulse control, and inappropriate movement and touch. The
104 sensory overresponsivity and inattentive (ADHD) subtypes are both characterised by
105 distractibility and difficulty focusing. Finally, the sensory underresponsivity and inattentive
106 (ADHD) subtypes both include being unaware when spoken to or when asked to follow
107 directions (Miller et al., 2012). However, studies comparing children with a dual diagnosis of
108 sensory modulation difficulties and ADHD with children with only one of these disorders
109 have shown that sensory modulation difficulties and ADHD behaviours can be reliably
110 distinguished from each other based on both parent-reports and behavioural measures (Ben-
111 Sasson et al., 2017; Reynolds et al., 2010; Yochman et al., 2013).

112 In children with ADHD, sensory modulation difficulties have been found to affect play,
113 social participation, and increased functional impairment (Cosbey et al., 2010;; Mangeot et
114 al., 2001; DeSerisy et al, 2019). Sensory modulation difficulties in children with ADHD can
115 be related to increased delinquent and aggressive behaviours (Mangeot et al., 2001; Ben-
116 Sasson et al., 2017) . Early detection and management of sensory difficulties can help to
117 improve functioning in children with ADHD (Ghanizadeh, 2011). As such, it is particularly
118 important to attempt to identify and treat sensory modulation difficulties in this population
119 (Wilkes-Gillan et al., 2016). Given that these sensory difficulties can impact abilities in
120 behavioural, emotional, motor, and cognitive domains, it is important to identify potential
121 difficulties at an early stage (Jorquera-Cabrera et al., 2017). Previous research has shown that
122 children who had both ADHD symptoms and sensory overresponsivity could be separated
123 from typically developing children by higher sensitivity scores at 18 months (Ben-Sasson et
124 al., 2017). This study was conducted with a community sample, used a dimensional approach,
125 and focused only on auditory and tactile processing. Additional studies investigating sensory

126 modulation across all sensory domains, and using both dimensional and diagnostic
127 approaches, can provide greater clarity on the development and aetiology of the disorder
128 (Dallos et al., 2017). Further evidence suggests that children with ADHD show sensory
129 sensitivity in infancy before attention, impulsivity, or hyperactive symptoms emerge (Kaplan
130 et al., 1994). Thus, exploring sensory modulation in children at-risk of ADHD may inform
131 whether early sensitivity is a risk marker for ADHD symptoms at school-age.

132 **1.2 ADHD and negative affect**

133 Putnam and colleagues have identified three factors of infant temperament: 1) surgency, 2)
134 negative affect, and 3) effortful control (Putnam et al., 2006). Negative affect is characterized
135 by responses such as fear, anxiety, discomfort, and anger or frustration (Allan et al., 2013).
136 Recent research indicates that markers of ADHD symptoms could be observed in early
137 development, through parent-reported child temperament (Frick et al., 2019; Miller et al.,
138 2020). In particular, it has been theorised that high negative affectivity in early development
139 may disrupt the development of effortful control, and by extension, act as an early marker for
140 ADHD (Sullivan et al., 2015).

141 A significant proportion of children with ADHD have been found to experience emotional
142 challenges such as poor emotional self-regulation, reduced empathy, and increased emotional
143 reactivity to negative environmental events (Okado et al., 2016). A study of 66 male infants
144 at familial risk of ADHD showed more anger reactivity at 7 months compared to typically
145 developing controls (Auerbach et al., 2004). Similarly, 6-month-old infants with a family
146 history of ADHD reported higher levels of distress than infants without a family history of
147 the disorder (Sullivan et al., 2015). High negative affect was associated with ADHD
148 symptoms of inattention, hyperactivity, and impulsivity in 3-6-year-old children (Martel et
149 al., 2014). Martel and colleagues (2014) suggest that negative affect may be particularly
150 relevant during early development when neural circuitry is rapidly growing. Similarly,
151 Wichstrom and colleagues (2018) observed that ADHD symptoms were predicted by high
152 negative affect in a Norwegian sample of children.

153 Nigg et al. (2004) report that externalizing behaviour problems including conduct disorder
154 and aggression can occur in more than 50% of children with ADHD. High negative affect is
155 associated with oppositional defiant disorder (ODD) (Eisenberg et al., 2009; Okado et al.,
156 2016). A similarly high co-morbidity rate has also been observed between ADHD and

157 psychiatric disorders, many of which are characterized by symptoms of negative mood such
 158 as depression and anxiety (Lonigan et al., 2003). Examining early emotional reactivity in
 159 children with a family history of ADHD may be one route to further understanding the
 160 disorder, particularly more complex cases.

161 **1.3 Sensory modulation and negative affect**

162 Deficits in sensory modulation have been associated with difficult temperament (Case-Smith
 163 et al., 1998; DeSantis et al., 2004). Infant studies have observed a relationship between
 164 sensory processing and negative affect (DeSantis et al., 2004; DeGangi et al., 1991). Most
 165 often, sensory hyperresponsivity has been positively correlated with aspects of negative affect
 166 such as fear and irritability (Keuler et al., 2011). One study reports that tactile, visual, and
 167 auditory defensiveness, all of which are aspects of hyperresponsivity, were significantly
 168 related to irritable temperament in preterm infants (Case-Smith et al., 1998). Infants
 169 described as being fussy, irritable, and having poor attention skills were found to also present
 170 with hyperresponsivity to tactile, visual, and vestibular stimuli (DeGangi et al., 1991). A twin
 171 study of young children observed moderate correlations between negative affect and fear with
 172 auditory and tactile hyperresponsivity (Keuler et al., 2011). A similar result was observed by
 173 Goldsmith and colleagues (2006), in which low to moderate positive correlations were found
 174 between sensory hyperresponsivity and various temperament scales such as sadness, anger,
 175 and soothability. Fear showed a relatively high correlation with hyperresponsivity (Goldsmith
 176 et al., 2006). Sensory hyperresponsivity has been observed as a risk factor for anxiety
 177 disorders, with hyperresponsive preschool children also displaying high levels of anxiety
 178 symptoms at school age (Carpenter et al., 2019). Early sensory hyperresponsivity has also
 179 been found to predict later anxiety in autistic children (Green et al., 2012). Treatments for
 180 sensory responsivity has been successful in decreasing anxiety in adults and young autistic
 181 children (Pfeiffer & Kinnealey, 2003; Baranek et al., 2015). Given the high co-occurring rate
 182 of anxiety and ADHD, (Reynolds & Lane, 2009) investigating whether sensory responsivity
 183 is contributing to this relationship could inform treatment and intervention options for this
 184 population. In the current study, we expect that higher levels of negative affect will be
 185 associated with higher levels of sensory hyperresponsivity.

186 In contrast to the body of literature examining hyperresponsivity and temperament, little
 187 focus has been paid to the relation between both sensory seeking and sensory

188 hyporesponsivity and negative affect. One study in an ASD population found an association
189 between both hyporesponsivity and sensory seeking patterns and more negative mood (Brock
190 et al., 2012). As such, within an at-risk ADHD population, it is unclear whether a relation
191 exists between the hyporesponsivity and negative affect, and which direction this relationship
192 might be. Similarly, it is unclear whether a relation exists between sensory seeking and
193 negative affect in this population.

194 To our knowledge, no empirical studies have examined the relation between negative affect
195 and sensory modulation in children at familial risk of ADHD. This study considers how
196 sensory modulation is associated with early temperament markers of ADHD in children at
197 familial risk for the disorder.

198 **1.4 The current study**

199 The current study aims to examine sensory modulation in children between the age of 7 and
200 72 months with a higher likelihood of developing ADHD due to familial diagnosis. This age
201 range was chosen as it is old enough to capture the temperament component of negative
202 affect (Putnam et al., 2008) yet younger than the typical age range of ADHD diagnosis at 6
203 to 12 years of age (Visser et al., 2014; Miller et al., 2020). A secondary aim of this study is to
204 explore the relation between sensory modulation, familial risk of ADHD, and the
205 temperament dimension of negative affect. While previous studies have reported a link
206 between poor sensory modulation and negative affect, no study to date has looked at this
207 relationship in children at-risk of later ADHD diagnosis. Understanding this relation between
208 negative affect and sensory modulation in children at-risk of ADHD is important to identify
209 whether a certain population of children (i.e. those with sensory processing problems and
210 negative emotional reactivity) could be at greater risk of developing later ADHD. Based on
211 previous research, the following research questions were proposed:

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

217 Research question 2: Is there an association between sensory modulation, familial risk of
218 ADHD, and negative affect? It is expected that children with greater levels of

219 hyperresponsivity will also have high levels of negative affect. In terms of
 220 hyporesponsiveness and sensory seeking, it is unclear which direction the relationship will be
 221 in, if any relationship does exist.

222 **2. Method**

223 **2.1 Participants**

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

235 Twenty one children were removed from analysis due to not meeting the inclusion criteria: 14
 236 were born preterm (n=4 high-risk, n=10 low-risk); 2 due to an epilepsy diagnosis (n=1 high-
 237 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-
 238 risk, n=1 low-risk), and 1 for being under 6 months (n=1 low-risk). One hundred and forty-
 239 five caregivers were included in the final analyses. Parents were asked whether there was a
 240 confirmed diagnosis of ADHD in the family and to indicate which family member received
 241 this diagnosis. Children were then divided into a high-risk or low-risk group based on family
 242 history of ADHD. Those in the high-risk group (n=47) had a history of ADHD in the
 243 immediate family (i.e. a parent (29.8%), sibling (57.42%) or both parent and sibling (12.8%)
 244 had an existing diagnosis). The low-risk group (n=98) had no immediate family history of
 245 ADHD. Children’s ages ranged from 7 months to 6 years 0 months (M= 2.74 months,
 246 SD=1.64). In the high-risk group, of the 18 families with co-occurring ASD and ADHD, 14
 247 had a sibling diagnosis of both ADHD and ASD, three had parents with a dual diagnosis, and
 248 one case where both a parent and sibling had ASD and ADHD. See Table 1 below for a
 249 demographic breakdown of the sample.

250 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	41(87.2%)	90(92.8%)	.403
	White			
	Mixed race	5(10.6%)	8 (6.6%)5(5.2%)	
	Other	1(2.1%)	2(2.1%)	
Single Parent?	Yes	10(21.3%)	4(4.1%)	.009
Maternal Education	Junior	4(11.1%)	2(2.2%)	.003
	Cert/GCSE			
	Leaving	5(13.9%)	2(2.2%)	
	Cert/A Levels			
	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

251

252 **2.2 Measures**

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

256 **2.2.1 Sensory Processing**

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

268 “at risk” for sensory processing difficulties. Scores greater than 2 standard deviations above
269 the mean are considered to be in the “deficient” range for sensory processing.

270 **2.2.2 Temperament**

271 **The Infant Behaviour Questionnaire (Very Short Form) (IBQ), the Early Childhood**
272 **Behaviour Questionnaire (Very Short Form) (ECBQ) and the Childhood Behaviour**
273 **Questionnaire (Very Short Form) (CBQ) were used to measure temperament.** The IBQ-
274 VSF is a caregiver report measure designed for use with children from 3 to 12 months of age,
275 the ECBQ-VSF is designed for use with children from 18 to 36 months of age and is also
276 recommended for use for children between 13 to 17 months of age (Putnam & Rothbart,
277 2006). The CBQ-VSF is the older version of the form designed for 3 to 8-year-old children
278 (Putnam & Rothbart, 2006). All three measures are formed of three subscales of surgency,
279 effortful control, and negative affect. The reliability of negative affect subscales across the
280 measures ranged from .70-.78. Scores on the negative affect subscale from the IBQ, ECBQ
281 and CBQ were transformed into Z-scores in order to compare across groups. Negative affect
282 has been shown to be generally stable across development (Putnam et al., 2008; Carranza et
283 al., 2013). The negative affect subscale contains questions such as how a child reacts to
284 unfamiliar persons, their frustrations, and soothability.

285 **2.3 Procedure**

286 Ethical approval for the study was obtained from BLINDED FOR REVIEW. All parents
287 provided informed consent before beginning the experiment. Questionnaires were completed
288 online using Qualtrics software (Qualtrics, 2005). Data collection commenced in February
289 2018 and was completed in March 2019. Participants first provided demographic information
290 before completing the IBQ, ECBQ or CBQ (depending on the age of the child) and the SEQ.
291 Some parents did not complete all questionnaires (81.3% of participants completed the SEQ
292 only). Those who did not complete the temperament measure did not differ from those who
293 did on maternal education, marital status, or family size. This study formed part of a larger
294 study investigating a range of potential early markers for ADHD in children at familial risk of
295 diagnosis.

296 **2.4 Statistical Analysis**

297 All statistical analyses were conducted using SPSS 24 (SPSS Inc.). To address the first
 298 research question, independent t-tests were carried out between the high-risk and low-risk
 299 groups to investigate differences in sensory processing patterns and sensory processing
 300 domains. Effect sizes for significant differences were reported with Cohen's d, where 0.2 =
 301 small effect, 0.5 = medium effect, 0.8 = large effect (Cohen, 1988). An independent t-test was
 302 also carried out between the high-risk and low-risk groups for the negative affect subscale of
 303 the temperament measure. To address the second research question, correlations were carried
 304 out between sensory processing patterns and negative affect. A hierarchical regression
 305 analysis was then conducted to determine whether sensory processing patterns and group
 306 membership predict negative affect. For the regression model, there was independence of
 307 residuals, as measured by a Durbin-Watson statistic of 2.021. Homoscedasticity was present,
 308 as assessed by visual inspection of a plot of studentized residuals versus unstandardized
 309 predicted values. There was no evidence of multicollinearity, with all tolerance values greater
 310 than 0.1, with no correlations higher than 0.8 (Berry et al., 1985; Neter et al., 2004). There
 311 were no studentized deleted residuals greater than ± 3 standard deviations, no leverage values
 312 greater than 0.2, and values for Cook's distance above 1. The assumption of normality was
 313 met, as assessed by a Q-Q Plot. Variables were added in three stages. Age as a continuous
 314 variable was added first to control for age-related differences. The second variable added to
 315 the model was group membership, as we expect that children with a diagnosis of ADHD in
 316 their family will show higher levels of negative affect in each respective model.
 317 Hyperresponsivity and hyporesponsivity scores from the SEQ were then added to the
 318 regression in the final stage.

319 **3. Results**

320 **3.1 Reliability analysis**

321 Reliability analysis was carried out on all scales. The IBQ, ECBQ, and CBQ had moderate
 322 reliability, with Cronbach's alpha of .687, .690, and .633 respectively. The SEQ had the
 323 strongest internal reliability, with Cronbach's alpha of .886.

324 **3.2 Sensory processing**

325 An independent t-test was carried out between the high-risk and low-risk groups to determine
 326 whether the groups were significantly different in relation to sensory processing patterns
 327 (Table 2). Results showed a significant difference between the groups in terms of SEQ

328 hyporesponsiveness ($p = .038, d = 0.39$) and SEQ hyperresponsiveness ($p = .041, d =$
 329 0.39). No difference was observed for the sensory seeking pattern or SEQ total score. As
 330 there were differences observed for two of the three sensory patterns, specific domains were
 331 explored, and no significant differences emerged. A significant portion of both groups scored
 332 greater than 1 standard deviation from the norm for overall sensory processing score (high-
 333 risk: 34.0%; low-risk: 27.5%). For hyporesponsiveness, the high-risk group had significantly
 334 more children scoring outside the normal range (55.2%) compared to the low-risk group
 335 (29.6%). For hyperresponsiveness, 31.9% scored outside the normal range compared to 16.3%
 336 of low-risk children, however this difference was not significant. More children from the
 337 low-risk group than the high-risk group scored in the ‘at-risk’ or ‘deficit’ categories for the
 338 sensory seeking pattern, indicating scores greater than 1 standard deviation from the mean
 339 (33.6% and 27.5% respectively), although this difference was not statistically significant. In
 340 all sensory patterns except sensory seeking, children from the high-risk group reported higher
 341 scores, indicating that some children at-risk of ADHD experience more extreme patterns of
 342 sensory processing.

343 Correlations were also carried out for the subscales of the SEQ (see Figure 1), as previous
 344 research has suggested that children can show varying levels of all three patterns (Brock et
 345 al., 2012). Results showed significant correlations between all three subscales:
 346 hyperresponsiveness and hyporesponsiveness ($r = .581, p < .001$); hyperresponsiveness and
 347 sensory seeking ($r = .474, p < .001$); and hyporesponsiveness and sensory seeking ($r =$
 348 $.356, p < .001$). 21.3% of children from the high-risk group are showing high levels of
 349 hyperresponsivity, hyporesponsivity, and sensory seeking. In contrast, only 3.1% of the low-
 350 risk group had elevated scores across all three patterns.

351 Figure 1: Bubble plot of hyporesponsive, hyperresponsive, and sensory seeking scores for
 352 high-risk group HERE

353 Figure 2: Bubble plot of hyporesponsive, hyperresponsive, and sensory seeking scores for
 354 low-risk group HERE

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

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

Variable	High-risk M (SD) N = 47	Low-risk M (SD) N = 98	<i>t</i>	<i>p</i>	<i>d</i>
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

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

361 **3.3 Negative affect**

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

364 **3.4 Associations between sensory processing patterns and negative affect**

365 For the whole sample, significant correlations were observed between negative affect and the
 366 overall SEQ score ($r = .316, p = .001$), the hyperresponsiveness subscale ($r = .382, p <$
 367 $.001$), and the sensory seeking subscale ($r = .201, p = .031$). There was a trend towards
 368 significance for the hyporesponsiveness pattern ($r = .181, p = .052$).

369 For the high-risk group, a significant correlation was observed between negative affect and
 370 total SEQ score ($r = .545, p < .001$), the hyperresponsiveness subscale ($r = .593, p <$
 371 $.001$), the hyporesponsiveness subscale ($r = .364, p = .027$), and the sensory seeking
 372 subscale, ($r = .432, p = .008$). In contrast, no significant correlations emerged between
 373 negative affect and sensory scores for the low-risk group. A trend towards significance was
 374 observed between negative affect and the hyperresponsiveness subscale for the low-risk
 375 group ($r = .214, p = .059$).

376 A hierarchical regression was carried out to determine whether sensory processing patterns
 377 and group membership could predict negative affect, when controlling for age across the

378 whole sample (Table 3 below). Age was controlled for as the two groups were not matched
 379 for age. The addition of group membership did not lead to a statistically significant change in
 380 R^2 . The full model of age, group membership, SEQ hyperresponsivity, SEQ
 381 hyporesponsivity was statistically significant, with an increase in R^2 of .127, $F(4,106) =$
 382 $6.008, p < .001, R^2 = .185, \text{adjusted } R^2 = .154$. Taken together, all of the variables
 383 accounted for 18.5% of variance in negative affect. Hyperresponsiveness was the only
 384 significant contributor to the model ($\beta = .455, p = .000$). Higher levels of hyperresponsiveness
 385 were associated with higher negative affect.

386

387 Table 3: Hierarchical regression of age, group membership, and sensory processing
 388 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**

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

390 **4. Discussion**

391 This study examined differences in sensory modulation between children with and without a
 392 familial history of ADHD. Findings showed that children at high-risk of ADHD were more
 393 likely to show both a hyperresponsive and hyporesponsive pattern of sensory processing,
 394 relative to children at low-risk of ADHD. This study is the first to report a higher rate of
 395 sensory modulation difficulties, which have already been established to be salient for children
 396 with ADHD, in young children at-risk of a later ADHD diagnosis due to familial history. A
 397 significant difference in sensory hyperresponsiveness between high-risk and low-risk
 398 children is concurrent with the literature on children with a confirmed diagnosis of ADHD. In
 399 the current study, 31.9% of children at-risk of ADHD reported at-risk/deficient scores of
 400 hyperresponsiveness. Reynolds et al. (2010) observed that 13 of their 24 participants
 401 (54.17%) with ADHD also had sensory hyperresponsivity. Similarly, Ben-Sasson et al.

402 (2017) report that 48% of a community sample who met the criteria for an ADHD diagnosis
 403 also met the criteria for hyperresponsivity. One study in an adult ADHD population report
 404 that 19% of participants were hypo-responsive, while further 9.5% of participants met the
 405 criteria for both hypo-responsiveness and hyperresponsiveness (Bijlenga et al., 2017). In the
 406 current study, a large portion of high-risk children (55.2%) reported extreme patterns of
 407 hypo-responsiveness (falling into the at-risk or deficient range), and 29.8% of children at-risk
 408 for ADHD showed extreme levels of both hypo-responsiveness and hyperresponsiveness. In
 409 comparison, only 7.1% of children from the low-risk group displayed extreme levels of both
 410 hypo-responsiveness and hyperresponsiveness, while 29.6% displayed at risk/deficient scores
 411 for hypo-responsiveness only. This is a large proportion of children (almost 30%) showing at-
 412 risk/deficient scores for hypo-responsiveness and it may be that the SEQ overestimates risk.
 413 Parents who suspected sensory processing difficulties in their children may also have been
 414 more motivated to take part in the study, and this may explain the elevated
 415 hypo-responsiveness scores.

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

424 An association was observed between hyperresponsivity and negative affect, similar to
 425 findings from previous research (DeGangi et al., 1991; Goldsmith et al., 2006). Child's age
 426 and familial risk for ADHD was not related to negative affect, while hyperresponsivity
 427 accounted for 18.5% of variance in negative affect levels. According to Cohen (1988), these
 428 adjusted R^2 values indicate a small to moderate effect size. Results indicate that higher levels
 429 of hyperresponsiveness predict higher levels of negative affect. This differs from a similar
 430 study examining the relation between sensory processing and temperament in a population
 431 with autism spectrum disorder (ASD). Brock and colleagues (2012) reported significant
 432 effects of association between a combination of sensory features (across all three patterns)
 433 and increased negative mood. A significant association was observed between

434 hyporesponsivity and slowness to adapt, low reactivity, and low distractibility. No significant
 435 main effects were found between hyperresponsiveness or sensory seeking and the
 436 temperament subscales (Brock et al., 2012).

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

447 **4.1 Limitations**

448 Certain limitations need to be taken into account when considering the current findings. The
 449 SEQ was developed for an autism population and related developmental disorders (Baranek,
 450 2018). As a result, this measure has not been validated prior to this study for use with a
 451 population at-risk of ADHD. Future research should further investigate the valid use of the
 452 SEQ in an ADHD population, and its psychometric properties. Furthermore, a significant
 453 portion of the high-risk group (38.3%) also had a family history of ASD. This could be a
 454 confounding factor as hyporesponsiveness and hyperresponsiveness have been well
 455 established in the ASD phenotype (APA, 2013). The results of this study found no difference
 456 between those with a family history of both ASD and ADHD and those with ADHD only,
 457 suggesting that sensory modulation difficulties are salient for children at-risk of ADHD.
 458 Further research using larger samples of children with a history of ADHD only, those with a
 459 history of ADHD and ASD, and children with no family history of either disorder are
 460 required to further elucidate this overlap. Sensory modulation difficulties may explain part of
 461 the shared phenotype between ADHD and ASD (Dellapiazza et al., 2020). Given that sensory
 462 modulation difficulties are also present across a range of neurodevelopmental disorders (e.g.
 463 Developmental Coordination Disorder; Delgado-Lobete et al., 2020) further research into
 464 early sensory processing may form part of the explanation for the underlying etiology of
 465 atypical development.

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

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

491 **4.2 Future research and implications**

492 Due to the current findings which show that sensory modulation difficulties can be present in
 493 children at familial risk of ADHD, sensory functioning should be considered in the evaluation
 494 of children who are at risk of ADHD (DeSerisy et al., 2019). Although, it is important to note
 495 that not all of the children in the high-risk group will later receive an ADHD diagnosis.
 496 However, it seems that a significant portion of children with ADHD do present with sensory
 497 modulation difficulties, and this subgroup may require a certain intervention approach

498 (Ghanizadeh, 2011). Early detection and treatment of these difficulties can help children with
499 ADHD in terms of their participation in social activities or prevent the development of
500 secondary social-emotional problems, such as anxiety (Yochman et al., 2004; Reynolds &
501 Lane, 2009). Previously, sensory interventions have been successful in reducing anxiety (an
502 element of negative affect) in typical adults and autistic children (Pfeiffer & Kinnealey, 2003;
503 Baranek et al., 2015). If early sensory modulation difficulties could be treated through
504 targeted interventions, this might reduce negative affect levels in children and in turn, reduce
505 later ADHD symptomatology. Understanding early sensory processing and the aetiology of
506 sensory modulation is an important avenue for future research and early intervention
507 planning.

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

517 **4.3 Conclusion and implications**

518 Previous research has shown that children with an ADHD diagnosis are likely to experience
519 extreme patterns of sensory processing that can interfere with daily life and exacerbate the
520 functional impairments of ADHD (Ben-Sasson et al., 2017; Mimouni-Bloch et al., 2018). To
521 the author's knowledge, this study is the first to demonstrate that these extreme patterns of
522 hyperresponsivity and hyporesponsivity are present in children at familial risk of ADHD.
523 This study also found that higher levels of hyperresponsivity were predictive of higher levels
524 of negative affect in this cohort. Recent research has identified the temperament domain of
525 negative affect as a potential early marker to later ADHD symptomatology (Auerbach et al.,
526 2008; Sullivan et al., 2015). As a result, it is important to consider whether young children
527 showing high levels of hyperresponsivity may be particularly vulnerable to later ADHD
528 diagnosis. This is an important potential avenue for early intervention, as treating early

529 sensory modulation difficulties could reduce negative affect levels and severity of later
530 ADHD symptomatology. Further research in this population is required to establish whether
531 sensory modulation difficulties are a precursor to ADHD symptomatology.

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536

537

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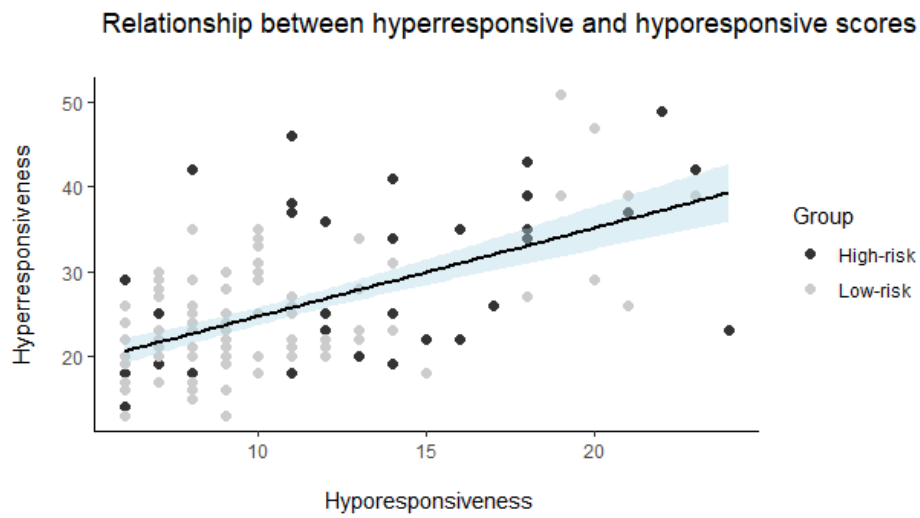
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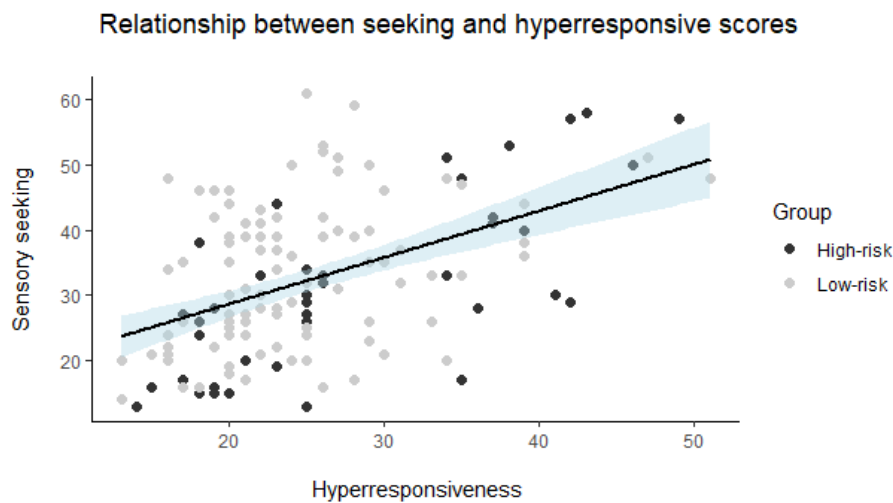
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811 **Appendix A: Scatterplots of SEQ subscale scores**



812

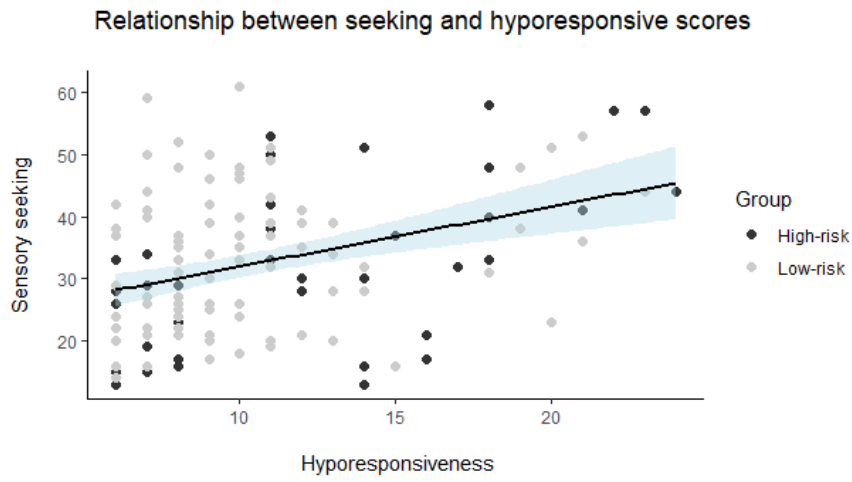
813 Figure A.1: Scatterplot of relationship between hyperresponsive and hypo-responsive scores
814 on the SEQ



815

816 Figure A.2: Scatterplot of relationship between hyperresponsive and sensory seeking scores
817 on the SEQ

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820 Figure A.3: Scatterplot of relationship between hyporesponsive and sensory seeking scores
821 on the SEQ

822