1	Running title: Sensory sensitivity and food fussiness in neurodevelopmental disorders
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3 The relationship between sensory sensitivity, food fussiness and food preferences in4 children with neurodevelopmental disorders

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- 20
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### 27 Abstract

28 Heightened sensitivity to sensory information has been associated with food fussiness 29 in both atypical and typical development. Despite food fussiness and sensory 30 dysfunction being reported as common concerns for children with neurodevelopmental 31 disorders, the relationship that exists between them, and whether they differ between 32 disorders, has yet to be established. The current study aimed to examine sensory 33 sensitivity as a predictor of food fussiness in three different neurodevelopmental 34 disorders, whilst controlling for comorbidity amongst these disorders. Ninety-eight 35 caregivers of children with Attention Deficit Hyperactivity Disorder (ADHD; n=17), 36 Tourette Syndrome (TS; n=27), Autism Spectrum Disorder (ASD; n=27), and typical 37 development (TD; n=27) were compared using parental reports of child food fussiness, 38 food preferences and sensory sensitivity. Children with neurodevelopmental disorders 39 were reported to have significantly higher levels of both food fussiness and sensory 40 sensitivity, with children with ASD and TS also showing significantly less preference 41 for fruit than children with TD. Importantly, higher levels of taste/smell sensitivity 42 predicted food fussiness for all four groups of children. In addition, taste/smell 43 sensitivity fully mediated the differences in food fussiness between each group of 44 neurodevelopmental disorder compared to the TD group. The findings highlight that 45 food fussiness is similar across these neurodevelopmental disorders despite accounting 46 for comorbidity, and that greater sensitivity to taste/smell may explain why children 47 with neurodevelopmental disorders are more likely to be fussy eaters.

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Keywords: Tourette syndrome; Attention Deficit Hyperactive Disorder; Autism
Spectrum Disorder; food fussiness; sensory sensitivity

## 51 **1. Introduction**

52 Neurodevelopmental disorders comprise a broad range of conditions emerging in 53 childhood and are primarily associated with impairments in the development of the 54 central nervous system and the brain (Posthuma & Polderman, 2013). These can 55 encompass a range of aetiologies, including a known mutation on a single gene (Fragile 56 X syndrome), disorders with polygenetic or unknown genetic origins (e.g. Attention 57 Deficit Hyperactivity Disorder [ADHD], Autism Spectrum Disorder [ASD], Tourette 58 Syndrome [TS]), and disorders from early/perinatal factors (e.g. cerebral palsy; Cascio, 59 2010). Population-based twin studies have shown high genetic correlations between

ASD, tic disorders and ADHD (Lichtenstein, Carlström, Råstam, Gillberg &
Anckarsäter, 2010), and it is this group of disorders which formed the focus of the
current study.

63 Each of these three neurodevelopmental disorders, as defined in the DSM-5, are 64 characterised by a distinct set of diagnostic features, such that TS is characterised by 65 involuntary, repetitive and non-rhythmic motor and vocal tics, ADHD by excessive and impairing inattentive, hyperactive and impulsive behaviour, and ASD by language 66 67 development, social and communication deficits (American Psychiatric Association, 68 2013). Despite this, these disorders have also been recognised as sharing many 69 overlapping features, in addition to being highly comorbid with each other. For 70 example, ADHD is diagnosed in 60% of individuals with TS (Freeman et al., 2000). 71 Recent studies have also documented that TS is comorbid with ASD (Cath & Ludolph, 72 2013), with research showing the presence of autistic symptoms in two thirds of 73 individuals with TS (Kadesjö & Gillberg, 2000), and the relationship between autism 74 symptomology in TS is particularly strong for individuals with early onset of TS 75 (Zappella, 2002). Furthermore, it is estimated that 50% of individuals with ADHD will 76 meet the criteria for diagnosis of ASD (Kochhar et al., 2011). Importantly in the context 77 of the current study, individuals with these disorders have also all been identified as 78 showing difficulties with feeding and eating concerns which extend beyond the 79 developmental stages of childhood, in which feeding and eating problems are very 80 common (Johnson, 2014). This raises the possibility that the comorbidity of these 81 disorders may explain their similarities in eating outcomes.

82 More specifically, the two eating outcomes which form the focus of this paper are food 83 preferences, understanding the likings of different food groups, and children's 84 behaviour towards food in terms of fussiness. Food fussiness can be defined as 85 consuming 'an inadequate variety of foods' (Galloway, Fioritio, Lee & Birch, 2005, p. 86 542). Caregivers commonly report food fussiness in children with typical development, 87 with around 46% of children reported as being picky eaters at some point between 1.5 88 and 6 years (Cardona-Cano et al., 2015). Although picky/fussy eating are relatively 89 stable traits (Mascola et al., 2010), a substantial proportion of children show reductions 90 in picky eating by 6 years (Cardona-Cano et al., 2015). In contrast, food fussiness is 91 more frequent and persistent in children with neurodevelopmental disorders (Bandini et al., 2017; Suarez, Atchison & Lagerwey, 2014). For example, food fussiness has 92

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93 been found to be one of the most commonly reported feeding problems in children with 94 ASD (Williams, Field & Seiverling, 2010), with a narrow range of foods being selected 95 based on food type, temperature, texture and colour (Seiverling, Williams & Sturmey, 96 2010). This is important, given the emotional and health implications of food fussiness 97 (Dovey, Staples, Gibson & Halford, 2008). Research has identified fussy eaters to 98 consume fewer foods containing vitamin E, C and fibre, likely due to their low intake 99 of fruit and vegetable. A lack of nutrient-rich foods can lead to nutritional deficiencies, 100 including magnesium and iron deficiencies, as well as slower growth patterns (Xue et 101 al., 2015; Antoniou et al., 2016). Food fussiness has also been associated with 102 additional stress and frustration for the child and their families (Rogers et al., 2012; 103 Curtin et al., 2015), along with difficulty eating in social environments (Nadon et al., 104 2011). These various adverse effects highlight the need for interventions to increase 105 food intake, in terms of variety and healthy foods, in children with fussy eating.

106 For children with typical development, research has established several methods to 107 encourage children to reduce food fussiness and increase consumption of fruit and 108 vegetables. These include parent modelling, which can help increase consumption of 109 novel foods through observational learning, and repeated re-offering of food to help 110 familiarise the child with the foods until they are willing to try (Holley, Haycraft & 111 Farrow, 2015; Wardle, Carnell & Cooke, 2005). However, not only have children with 112 heightened levels of food fussiness been shown not to respond to these methods in the 113 same manner as typically developing (TD) children (Wolke, Schmid, Schreier & 114 2009), research addressing these interventions in children with Meyer, 115 neurodevelopmental disorders is limited.

116 Recommendations often given to parents, such as only presenting family meals instead 117 of the child's preferred foods, have found to be ineffective for children with ASD who 118 have feeding problems and may lead to severe nutritional deficiencies (Rogers et al., 119 2012). Behavioural interventions, such as behavioural momentum (Patel et al., 2007) 120 have been reported to be effective in reducing feeding problems and increasing food 121 consumption in children with ASD. However, these are single case studies and pre-post 122 intervention studies are needed to demonstrate the generalisability of treatment effects 123 (Matson & Fodstad, 2009). Ultimately, it is important to understand the underlying 124 aetiology of food fussiness in these clinical groups to devise the most effective 125 intervention strategy.

Given the comorbidity between TS, ASD and ADHD, it is important to understand 126 127 whether the underlying causes of food fussiness is syndrome specific or the shared 128 pathologies across the three neurodevelopmental disorders accounts for the high levels 129 of food fussiness evidenced in these groups of children (Smith, Rogers, Blissett & 130 Ludlow, 2019). One potential explanation for food fussiness is the associated 131 pathologies unique to the disorder. For example, some feeding difficulties may reflect 132 limited interests and difficulty in accepting change (Curtin et al., 2015). Evidence also 133 suggests that children with heightened motor impulsivity and reduced inhibitory control 134 and/or characteristics of ADHD weigh more. Children with ADHD are also more 135 susceptible to altered food intake patterns dependent on stressors, such as distress or 136 external cues including the time of day (Bennett & Blissett, 2017).

137 A common feature of all three neurodevelopmental disorders is an impairment in 138 sensory processing, which could provide an alternative explanation for food fussiness. 139 Impaired sensory processing may lead to over-responsiveness to stimuli (resulting in 140 more fussy eating), or under-responsiveness to stimuli (which may result in a desire for 141 more sweet, salty or fatty foods). Both sensory processing issues have been shown to 142 limit the range of food consumed and social enjoyment of eating (Johnson et al., 2014). Furthermore, in children with both typical and atypical development, the perceived 143 144 sensory properties of food are often considered to underlie children's reasons for rejecting food (Martins & Pliner, 2005). The focus of the current study is to explore 145 146 sensory over-responsiveness across these neurodevelopmental disorders in relation to 147 food fussiness.

148 Individuals with sensory over-responsiveness "respond to sensation faster, with more intensity, or for a long duration than those with typical sensory responsivity" (Miller, 149 150 Anzalone, Lane, Cermak & Osten, 2007, p.136). This increased sensitivity to sensory 151 information, such as taste, smell and touch, has been identified as an inherent 152 characteristic that makes one particularly vulnerable to be a fussy eater. For example, 153 a reluctance to eat new foods and/or eat fruit and vegetables has been associated with 154 higher levels of tactile and taste/smell sensitivity (Coulthard & Blissett, 2009). For 155 example, tactile oversensitivity has also been shown to have an impact upon the eating 156 habits and food selection in children with and without atypical development (Cermak, 157 Curtin & Bandini, 2010). High levels of taste/smell sensitivity (Tomchek & Dunn,

158 2007), and difficulty with texture have been associated with a lack of variety in the diet 159 in children with ASD (Schmitt, Heiss, & Campbell, 2008), and TS (Smith et al., 2019). 160 Importantly, oral sensitivity which also forms part of tactile sensitivity, has also been 161 associated with food fussiness in children with ASD and ADHD (Chistol et al., 2018; 162 Ghanizadeh, 2013). Greater prevalence of sensory sensitivities have been found in 163 children with ASD (Bizzell, Ross, Rosenthal, Dumont, & Schaaf, 2019; 164 Simpson, Adams, Alston-Knox, Heussler, & Keen, 2019), ADHD (Ghanizadeh, 2011; Lane, Reynolds, & Thacker, 2010), and TS (Belluscio, Jin, Watters, Lee & Hallett, 165 166 2011) compared to TD children. The relationship between sensory processing and food 167 fussiness in children with these neurodevelopmental disorders, without comorbidity 168 within these disorders, therefore warrants further investigation.

169 Despite the similarities in food fussiness found across these neurodevelopmental 170 disorders, there has been no research carried out directly comparing food fussiness across different neurodevelopment disorders and/or examining which factors are 171 172 particularly associated with eating difficulties in children without comorbid 173 neurodevelopmental disorders. The current study therefore aimed to be the first study 174 to directly compare food fussiness and food preferences, along with the role of sensory 175 sensitivity in predicting these eating outcomes, amongst children with ADHD, ASD, 176 TS and TD children. It was hypothesised that children with neurodevelopmental 177 disorders would display higher levels of food fussiness and sensory sensitivity than TD 178 children, and that levels of food fussiness would be comparable across each 179 neurodevelopmental disorder despite no other comorbid disorder being present. It was 180 also predicted that sensory sensitivity would be a predictor of eating outcomes for all 181 groups of children.

## 182 **2. Method**

#### 183 **2.1** Participants

One hundred and thirteen mothers reported information on their children between Spring 2017 and Autumn 2018. Children were screened for missing data (N = 6) and comorbidity for other neurodevelopmental disorders (N = 7; 2 TS were removed for having comorbidity with ASD, 1 TS with ADHD, 3 ASD with ADHD, and 1 TS with ADHD and ASD) by asking caregivers for their child's full diagnosis and whether they had been diagnosed with any additional disorders. Data from 98 mothers aged 25-67

- 190 years (M=40, SD=1) remained. Twenty-seven children had a diagnosis of TS, 27 had a
- 191 diagnosis of ASD, 17 had a diagnosis of ADHD, and 27 were typically developing
- 192 children with no known clinical diagnosis (5 females, 22 males) recruited through local
- 193 schools and forums. The groups did not differ in age, F(3,98) = .64, p = .59.
- 194 2.1.1 Children with Tourette syndrome

195 Twenty-seven children with a clinical diagnosis of TS (5 females, 22 males) were aged 196 between 6 years 7 months and 15 years 11 months. Caregiver report of a TS diagnosis 197 and the Premonitory Urge for Tics Scale (PUTS; Woods, Piacentini, Himle, & Chang, 198 2005) were used to confirm children's status in the TS group only. This measure reflects 199 the presence and frequency of premonitory urges, along with the relief that may be 200 experienced after tics have been performed, and is a tool used to estimate impact of 201 symptoms. A score above 31 indicates extremely high intensity with probable severe 202 impairments. In the current sample scores ranged from 9 to 35 (M=22, SD = 5). Of the children with TS taking medication (n = 13), the most commonly reported was 203 melatonin (n = 6). Other prescription drugs recorded were sertraline (n = 4) and 204 205 clonidine (n = 2).

## 206 2.1.2 Children with Autism Spectrum Disorder

207 Twenty-seven children with a clinical diagnosis of an ASD (11 females, 16 males) were 208 aged between 6 years 7 months and 17 years. Children with ASD were required to meet 209 the appropriate cut off on the Autism Spectrum Screening Questionnaire (ASSQ; 210 Ehlers, Gillberg, Wing, 1999). The ASSQ comprises 27 items rated on a 3-point scale, 211 0 indicating normal, 1 some abnormality and 2 definite abnormality. The range of score 212 is 0-54. Eleven items tap topics regarding social interaction, six cover communication 213 problems and five refer to restricted and repetitive behaviour. The remaining five items 214 measure motor clumsiness and other associated symptoms including motor and vocal 215 tics. All children reached the cut-off scores of 19 or more (M = 27, SD = 7), with mean 216 scores on subscales: Social Interaction (M = 11, SD = 7), Communication (M = 8, SD =3), Restrictive and Repetitive Behaviours (M = 4, SD = 2), and Motor skills and 217 218 clumsiness (M = 4, SD = 2). Of the children with ASD taking medication (n = 15), the 219 most commonly reported was melatonin (n = 6). Other prescription drugs recorded were 220 Prozac (n = 2) and clonidine (n = 2).

## 221 2.1.3 Children with Attention Deficit Hyperactive Disorder

222 Seventeen children with a clinical diagnosis of ADHD combined type. (7 females, 10 223 males) were aged between 6 years 2 months and 16 years 8 months. All of the children 224 met the required T-score of 65 or above on the Connors' Parent Rating Scale-Revised 225 (CPRS-R; Conners et al., 2008). Children's T-scores were reported as follows on the 226 content scales: Inattention (M = 85, SD = 7), Hyperactive/Impulsive (M = 86, SD = 6); 227 Learning Problems (M = 77.57, SD = 10.29), Executive Functioning (M = 84, SD = 10), 228 Aggression (M = 77, SD = 16), Peer Relations (M = 86, SD = 6), and for the symptom 229 scales: DSM-IV ADHD Inattention (M=84, SD = 9), DSM-IV ADHD Hyperactivity-230 Impulsive (M = 87, SD = 6), Conduct Disorder (M = 70, SD = 19), and Oppositional 231 Defiant Disorder (M = 79, SD = 12).

## 232 **2.2 Measures**

Demographic variables collected included: child's sex, birth date, any clinical diagnosis including comorbid disorders. Caregivers were asked to provide a measurement of their child's weight and height, which was then converted to a BMI standard deviation score (SDS). The Child Growth Foundation Package (1996) was used to standardise the measurements for age and sex according to standardised norms for a UK sample. Caregivers were also asked to describe their age, ethnicity and their relation to the child. Finally, all caregivers were asked to complete the following questionnaires:

## 240 2.2.1 The Short Sensory Profile (SSP; McIntosh, Miller, Shyu & Dunn, 1999)

241 The SSP is a 38-item an adapted version of the original Sensory Profile (Dunn, 1999) 242 designed to assess children's responses to sensory stimuli. The three subscales from the 243 questionnaire, which have been found to be common correlates of food fussiness, were 244 used to assess children's tactile sensitivity (e.g. avoids going barefoot, especially in 245 grass and sand), taste/smell sensitivity (e.g. avoids tastes or food smells that are 246 typically part of a child's diet), and visual/auditory sensitivity (e.g. covers eyes, or 247 squints to protect eyes from light). Caregivers responded to items on a 5-point Likert 248 scale ranging from 1 (always) to 5 (never) with lower scores indicating higher sensory 249 sensitivity. SSP total scores can range from a minimum of 38 (greatest frequency of 250 sensory symptoms) to 190 (no sensory symptoms). McIntosh et al., (1999) have shown 251 good psychometric properties internal consistency of the total and subscale scores 252 (Cronbach's alpha ranged from 0.68 to 0.92) with a discriminant validity of 95% in

distinguishing children with and without sensory modulation difficulties. In the current study good to excellent internal reliability was found for the subscales used; tactile sensitivity (Cronbach  $\alpha$ =.88), taste/smell sensitivity (Cronbach  $\alpha$ =.95), visual/auditory sensitivity (Cronbach  $\alpha$ =.90).

## 257 2.2.2 The Food Preference Questionnaire for children (FPQ; Fildes et al., 2015)

The FPQ requires caregivers to rate their child's liking for 75 commonly consumed 258 259 individual foods from 6 food groups: fruit, vegetables, meat/fish, dairy, snacks and 260 starches. Originally developed using data from a cohort of United Kingdom twins born 261 in 2007 Gemini study (n=2686), the food items on the questionnaire are rated on a 5-262 point Likert scale, ranging from 1 (dislikes a lot) to 5 (likes a lot), with an option of 263 'never tried' which is scored as a missing response. The mean score of items pertaining 264 to each subscale was calculated, with the higher the score indicating an increased like 265 towards the given food category. This measure has been used to understand the 266 children's food preferences longitudinally (Skinner, Carruth, Bounds & Ziegler, 2002) 267 and food preferences have been previously found to be a predictor of food consumption 268 (Drewnowski & Hann, 1999). In terms of psychometric properties, the current study 269 found a good to excellent internal reliability for the food groups; fruit (Cronbach  $\alpha$ =.95), vegetables (Cronbach  $\alpha$ =.93), meat/fish (Cronbach  $\alpha$ =.92), snacks (Cronbach 270 271  $\alpha$ =.82), dairy (Cronbach  $\alpha$ =.74), however the reliability for the starch subscale was 272 lower (Cronbach  $\alpha$ =.66).

# 273 2.2.3 The Children's Eating Behaviour Questionnaire (CEBQ: Wardle, Guthrie, 274 Sanderson & Rapoport, 2001)

The Children's Eating Behaviour Questionnaire (CEBQ) is a 35-item parent-report 275 276 questionnaire that assesses individual eating styles of children. The food fussiness 277 subscale of the CEBQ was used in the current study to assess parental perceptions of 278 their child's food fussiness behaviour (Sandvik, Ek, Eli Somaraki, Bottai & Nowicka, 279 2019). This subscale consists of six items and includes how difficult the child is to 280 please with meals; how often the child refuses to taste new foods subscale (food 281 neophobia) and the variety of foods the child will eat (picky eating). Caregivers rated 282 the frequency of which the child exhibits the behaviour on a 5-point Likert scale ranging 283 from 1 (never) to 5 (always). An average of the six food fussiness items was calculated. 284 A high score indicates that the child displays high levels of food fussiness. 285 Development of the questionnaire revealed good internal reliability coefficients

- 286 (Cronbach's alpha) for all the subscales, ranging from 0.74 to 0.91 (Wardle et al., 2001).
- 287 In the present study Cronbach's alpha for food fussiness was 0.68.

## 288 **2.3** *Procedure*

289 Ethical approval for this research was obtained from the University of Hertfordshire 290 University Ethical Advisory Committee Protocol Number: aLMS/PGT/UH/02784(4) 291 and the research was performed in accordance with the Declaration of Helsinki. 292 Participants were recruited through Tourettes Action, National Autistic Society charity 293 online website, online forums, local organisations, and mainstream and Special 294 Education Needs (SEN) schools who agreed to advertise the study. Participants 295 volunteered to partake by clicking on the given link, which directed them to the online 296 survey. The online participant information sheet provided further details about the 297 study, and those wishing to continue were required to provide informed consent by 298 signing an online consent form. Following this, every participant was presented with 299 the questionnaires in the same order. Information on how to seek further advice if the 300 parents had any concerns regarding their child's eating behaviours was also provided. 301 The survey took approximately 25 minutes to complete and was active for two months. 302 Families were provided no incentive to take part. At the end of the study, participants 303 were provided details of support groups for any concerns around difficulties in their 304 child's eating behaviours and reminded how to they could withdraw their data from the 305 study.

#### 306 **2. 4.** Analysis

307 A one-way ANOVA was first computed to compare differences in BMI SDS between 308 groups. Secondly, an independent t-test was conducted to examine whether there were 309 sex differences in outcome measures; Levene's test examined homogeneity of variance 310 and significance was reported appropriately. Subsequently, Two-tailed Pearson's 311 correlations were used to establish whether child age or BMI SDS were related to food 312 fussiness.

To investigate differences between the children with and without neurodevelopmental disorders, a series of one-way ANOVAs and post-hoc tests were conducted for each of the questionnaires (SSP, FPQ & CEBQ). To examine whether sensory sensitivity was a predictor of eating outcomes in the four groups (TS, ASD, ADHD & TD), a series of multiple linear regressions were carried with three of the sensory subscales (tactile,
taste/smell, visual/auditory) as a predictors of food fussiness.

319 Mediational analyses were used to evaluate differences between each group of 320 neurodevelopmental disorders compared to TD in relation to food fussiness, and to 321 examine whether sensory sensitivity mediated this relationship. Three separate 322 mediations were carried out using the procedure and macros provided by Preacher and 323 Hayes (2008). The effect of the group (coded 0 = TD, 1 = clinical group) was used as 324 an independent predictor of food fussiness in separate analyses for each clinical group, 325 including taste/smell sensitivity as a mediator of this relationship. Categorical data has 326 been shown to be appropriate to use as an independent variable in a mediation analysis 327 (Iacobucci, 2012). The recommendations of Hayes (2013) were followed, using dummy 328 coding to represent comparisons of interests and using the asymmetric bootstrap 329 Confidence Interval.

## **330 3. Results**

## 331 3.1. Descriptive statistics

332 A one-way ANOVA revealed no significant differences in BMI SDS between the four 333 groups of children, F(3, 73) = 1.05, p < .38. Across the total sample of children, two-334 tailed Pearson's correlations indicated that child food fussiness was not significantly associated with child age, r(99) = -.115, p = .26), or BMI SDS, r(75) = -.059, p = .61).<sup>1</sup> 335 336 An independent samples t-test revealed no significant difference in food fussiness 337 between males and females when comparing the total sample of children, t = (96) = .26, 338 p = .13. Therefore, these measures were not controlled for in further analyses. Two-339 tailed Pearson's correlations were also carried out to see if food fussiness was 340 associated with any of the symptom measures in children with neurodevelopmental 341 disorders. For children with TS, PUTS total was not significantly correlated with food 342 fusiness, r(24) = -.24, p = .26. For the children with ASD, the ASSQ total was not 343 significantly correlated with food fussiness, r(26) = .17, p = .41. For children with 344 ADHD, none of the subscales from Conner's parent rating scale significantly correlated

<sup>&</sup>lt;sup>1</sup> Food fussiness did not correlate with age or BMI SDS when split into diagnostic groups.

with food fussiness, including DSM-IV Inattention, r (15) = .28, p = .31, DSM-IV Hyperactivity, r (15) = .28, p = .38.

## 347 3.2. Differences in food fussiness, food preference and sensory sensitivity

To examine whether there were group differences in food fussiness, measured by the 348 CEBQ, a one-way ANOVA was conducted. The results revealed significant differences 349 350 between the groups on food fusieness, F(3, 97) = 6.29, p = .001. Tukey's HSD post hoc tests, as shown in Table 1, revealed children with TS (p = .004), children with ASD 351 (p = .001), and children with ADHD (p = .02) to have significantly higher levels of 352 food fussiness compared to children with TD. There were no significant differences in 353 354 food fussiness between the three different clinical groups (TS vs ASD p = .99; ADHD 355 vs TS p = .99; ADHD vs ASD p = .98). Mean scores and standard deviations are shown 356 in Table 1.

## 357 Table 1: Mean scores (standard deviation) for each of the questionnaires for children

with neurodevelopmental disorders and typically developing childr	·en.
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	TD (n=27)	TS (n=27)	ASD (n=27)	ADHD (n=17)
	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)
Demographics				
Age	9.7(2.4)	10.2(2.6)	10.4(3.2)	10.8(3.6)
Height	143.5(16.5)	146.7(17.6)	144.7(26.4)	147.8(28.0)
Weight	37.4(18.4)	39.4(17.8)	42.4 (19.2)	60.5(10.6)
BMI SDS kg/m <sup>2</sup>	5(1.9)	.6(4.1)	.9(1.3)	.6(2.2)
CEBQ				
Fussiness	3(1.0)	4(1.3)	4(1.02)	4(.9)
FPQ				
Meat/Fish	4(.5)	4(.8)	4(1.0)	4(.6)
Dairy	3(.6)	3(.8)	3(.8)	4(.8)
Starches	4(.6)	4(.7)	3(1.1)	3(.7)
Snacks	4(.7)	4(.6)	4(.5)	4(.6)
Fruit	4(.6)	3(1.0)	3(1.2)	4(.6)
Vegetables	3(.8)	3(1.0)	3(1.1)	3(.8)
Sensory Profile				
Tactile	32(5.0)	22(6.6)	23(6.7)	25(5.0)
Taste/Smell	18(3.1)	11(6.1)	11(5.5)	13(4.1)
Visual/Auditory	24(2.2)	16(5.9)	13(5.4)	19(5.0)
Overall	165(23.6)	114(30.2)	127(45.9)	112(20.9)

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360 To examine whether there were group differences in preference to food categories, as 361 defined by the FPQ, a series of one-way ANOVAs were conducted. The results

362 revealed significant differences between the groups on the following food categories: Starch, F(3, 97) = 4.97, p = .003, Fruit, F(3, 98) = 7.64, p < .001 and Vegetables F(3, 98) = 7.64, p < .001 and Vegetables F(3, 98) = 7.64, p < .001 and Vegetables F(3, 98) = 7.64, p < .001 and Vegetables F(3, 98) = 7.64, p < .001 and Vegetables F(3, 98) = 7.64, p < .001 and Vegetables F(3, 98) = 7.64, p < .001 and Vegetables F(3, 98) = 7.64, p < .001 and Vegetables F(3, 98) = 7.64, p < .001 and Vegetables F(3, 98) = 7.64, p < .001 and Vegetables F(3, 98) = 7.64, p < .001 and Vegetables F(3, 98) = 7.64, p < .001 and Vegetables F(3, 98) = 7.64, p < .001 and Vegetables F(3, 98) = 7.64, p < .001 and P = .000, 363 364 (98) = 3.41, p = .02. There were no significant differences for Meat, F(3, 96) = 2.32, p365 = .08; Dairy, F(3, 97) = .62, p = .61; Snacks, F(3, 97) = .77, p = .52. To further explore 366 the group differences in preference for starch and fruit and vegetables, post-hoc Tukey's HSD tests were conducted. Children with TS (p = .002) and children with ASD 367 368 (p < .001) had significantly lower preference for fruit than TD children. Children with ASD (p = .011) had significantly lower preference for vegetables than TD children. The 369 children with TS (p=.02) and TD children (p=.005) had a significantly greater 370 371 preference for starch than children with ASD. The remaining comparisons did not yield 372 significant differences in these food categories, and there were no differences in 373 preference for any food categories between children with ADHD and the other groups.

374 Finally, to examine whether there were group differences in sensory sensitivity, the 375 SSP total score and three selected subscales were analysed through a series of one-way 376 ANOVAs. The results revealed significant differences between the groups on overall 377 sensory sensitivity, F(3, 97) = 14.25, p < .001; taste/smell, F(3, 96) = 11.07, p < .001; 378 tactile F (3, 96)= 14.52, p < .001; and visual/auditory sensitivity, F (3, 97) = 23.29, p < .001379 .001. Compared to TD children, post-hoc Tukey's HSD tests revealed children with TS, 380 children with ASD and children with ADHD showed greater overall sensitivity (p 381 <.001), and greater sensitivity to the following: taste/smell (TS, ASD and ADHD p <382 .001), tactile (TS p < .001; ASD p < .001; ADHD p = .002), and visual/auditory 383 information (TS p < .001; ASD p < .001; ADHD p = .003). There were no significant 384 differences between the three neurodevelopmental groups on any of the four sensory 385 measures tested.

386 **3.3.** Multiple regressions

Multiple linear regression analyses were carried out to explore the relationship between the individual sensory subscales as predictors of food fussiness, and these were all entered into the model in the same step. As shown in table 2, taste/smell sensitivity was found to be the only significant predictor for food fussiness in all groups.

- 391 Table 2: Standard Coefficients of the three sensory profile subscales predicting food
- 392 *fussiness*

Tactile Taste/ Visual/ R<sup>2</sup> F

		Smell	Auditory		
TD					
Food Fussiness	02	64***	46	.45	7.99**
TS					
Food Fussiness	.11	81***	13	.63	15.43***
ASD					
Food Fussiness	.33	55*	47	.39	6.03*
ADHD					
Food Fussiness	.13	75*	.14	.27	2.95

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# *Note:* \*\*\* = *p* < .001, \*\* = *p* < .01, \* = *p* < .05

# 394 **3.3.** *Mediation analyses*

395 The findings from the previous multiple regression analysis revealed only taste/smell 396 sensitivity to be an independent predictor of food fussiness in all four groups of 397 children. Therefore, we addressed whether the taste/smell sensitivity subscale was also 398 an independent mediator of the relationship between food fussiness for each 399 neurodevelopmental group compared to the TD group. Separate multiple linear 400 regressions were used to explore the mediating role of taste/smell sensitivity on 401 differences between food fussiness for each neurodevelopmental disorder compared to 402 TD (e.g. TS vs TD, ASD vs TD, ADHD vs TD).

403 The difference in food fussiness between each neurodevelopmental group compared to 404 TD was found to be mediated by taste/smell sensitivity. As Figure 1 illustrates, the 405 standardized regression coefficient between group differences and taste/smell 406 sensitivity was statistically significant, as was the standardized regression coefficient 407 between groups and food fussiness. A significant Sobel test was found for each group analysis (TS vs TD, Z = -4.07, p < .001, ASD vs TD, z = -3.63, p < .0013, and ADHD 408 409 vs TD z = -3.02, p < .001) showing differences existed between each neurodevelopmental group compared to TD, and that their food fussiness was fully 410 411 mediated by the level of taste/smell sensitivity. These findings were confirmed using a 412 bootstrapping method with 5,000 resamples and 95% confidence intervals (Hayes, 413 2013). Results showed that the confidence interval did not include 0 in each of three separate analyses (TS vs TD LCI =-1.64: HCI = -.61; ASD vs TD LCI = -1.26: HCI = 414 -.38; ADHD vs TD LCI =-1.23: HCI = -.36), confirming the indirect effect was 415 statistically significant for each case. 416



417

418 Figure 1: Mediation model of group and taste/smell sensitivity on food fussiness

# 419 **4. Discussion**

The present study was the first to directly compare food fussiness across three different neurodevelopmental disorders, whist excluding comorbidity among these disorders. The findings failed to differentiate levels of food fussiness between TS, ASD and ADHD, demonstrating heightened food fussiness and sensory sensitivity for each disorder compared to TD children. Importantly, greater taste/smell sensitivity was found to mediate the differences in food fussiness between the TD group compared to the clinical groups, suggesting that it is greater sensitivity to sensory information in the
taste/smell domain which can account for why children with these neurodevelopmental
disorders have increased levels of food fussiness.

429 However, differences across the neurodevelopmental groups were also identified in 430 terms of food preferences. In contrast, to previous research showing males with ADHD 431 to consume less fruit and vegetables than TD children, the children with ADHD did not 432 present with any preferential differences to food categories. The research highlights the 433 need to further explore contextual factors in food preferences in children with ADHD 434 children. Only children with ASD were found to show a lower preference for vegetables 435 compared to children with TD. Consistent with previous research, children with TS and 436 children with ASD were found to show lower preference for fruit in comparison to 437 children who were TD (Smith et al., 2019; Maclin, Kandiah, Haroldson, & 438 Khubchandani, 2017). The lack of ADHD preference effect Increased food fussiness 439 and reduced fruit and vegetable preference, across TS and ASD, could lead to an 440 unvaried diet with many adverse health implications. Low consumption of plant-based 441 foods, including both fruit and vegetables, have been associated with an increased risk 442 of cardiovascular disease, obesity and diabetes (Slavin & Lloyd, 2012; Aune et al., 443 2017), and can lead to fatigue and deficiency in vital vitamins and minerals (Galloway 444 et al., 2005). This highlights the importance of exploring approaches to encourage 445 consumption of healthier foods, and the potential value of a focus on increasing fruit 446 and vegetable acceptance in children with neurodevelopmental disorders. However, 447 akin to existing literature on neurodevelopmental disorders, higher levels of food 448 fussiness were not found to be associated with children's BMI (Curtin, Bandini, Perrin, 449 Tybor & Must, 2005; Emond, Emmett, Steer & Golding, 2010).

450 Consistent with previous research, higher food fussiness was predicted by taste/smell 451 sensitivity in all groups of children (Cermak et al., 2010; Smith et al., 2019). This 452 provides further verification for the relationship between taste sensitivity and food 453 fussiness in children with both typical and atypical development (Cermak, et al., 2010). 454 However, unlike previous research that has suggested higher levels of tactile sensitivity 455 in ADHD (Ghanizadeh, 2013) and ASD (Schmitt et al., 2008) to be associated with 456 food fussiness, no significant relationship was established using the tactile measure, 457 although this may have been due to the small sample sizes. Instead, this is the first study to show that greater taste/smell sensitivity may account for differences in food fussinessbetween children with and without neurodevelopmental disorders.

460 The role of taste/smell sensitivity in food fussiness in children with these 461 neurodevelopmental disorders has important clinical implications, meaning pathways 462 of interventions should prioritise techniques which consider sensory sensitivity. 463 Repeated exposure techniques may be useful to gradually desensitise children and 464 increase their acceptance of different sensory experiences (Farrow & Coulthard, 2012). 465 Although, in children with neurodevelopmental disorders, such interventions may need 466 to be carried out over a lengthier time period than with TD children (Kim, Chung & 467 Jung, 2018), due to resistance to change food repertoire and unwillingness to try novel 468 foods identified in these clinical samples (e.g. Marí-Bauset, Zazpe, Mari-Sanchis, 469 Llopis-González, & Morales-Suárez-Varela, 2014). Additionally, children with ASD 470 have been found to explore foods for longer before making a hedonic decision than TD 471 children (Luisier et al., 2019). It is possible that providing children with 472 neurodevelopmental disorders with more time to explore their food could help them to 473 manage their sensory experiences independently and increase their familiarity with and 474 acceptance of exposed foods. In such cases, a multidisciplinary team, including 475 occupational therapists and registered dieticians, would be useful to individualise 476 interventions to the sensory characteristics of each child.

477 There are limitations of the current study that need to be noted. Firstly, while the food 478 preference questionnaire identified a reduced preference for fruit in the TS and ASD 479 groups, the absence of a food diary means specific detail on the types of foods children 480 with neurodevelopmental disorders eat, regarding both frequency and portion size, is 481 lacking (Day, McKeown, Wong, Welch & Bingham, 2001). The use of parent report 482 for height, weight and feeding problems has also been highlighted as not being the most 483 reliable method to gain information on children's BMI and feeding difficulties (for a 484 review see Arts-Rodas & Beniot, 1998). For example, parents may perceive even minor 485 feeding problems as major (Archer & Szatmari, 1990). There is a possibility that use of 486 parent-report for anthropometrics may also be discrepant to objective measures and 487 lead to miscalculation of BMI (Weden et al., 2013). Although objective measures are 488 optimal, where this is not feasible studies have highlighted strong and positive 489 correlations of parent-report of height and weight with objective measurements 490 (Haycraft & Blissett, 2012), and parents to be as accurate as a trained clinician in their

491 reporting (Chai et al., 2019). All children should also have been screened for other 492 neurodevelopmental symptomology; screening tools to assure correct inclusion in 493 diagnostic groups were used, but it is possible that screening all the children in the study 494 for each disorder of interest may have identified additional undiagnosed comorbid 495 problems. In the SSP, taste and smell sensory domains are combined into a single 496 subscale meaning there is an inability to differentiate between these characteristics 497 (Hubbard, Anderson, Curtin, Must & Bandini, 2014). Additionally, it is noted that there 498 are some items for the sensory taste/smell subscale that overlap with items for food 499 fussiness. Therefore, further work probably needs to make use of alternative measures 500 and observational sensory tests to confirm the importance and address the specific role 501 of sensory sensitivity in this domain for explaining food fussiness in children with 502 neurodevelopmental disorders.

503 The present study was the first to demonstrate that similar, high levels of food fussiness 504 are present across several individual neurodevelopmental disorders, thus indicating that 505 comorbid diagnoses do not underlie the effect, as previously suggested. It also suggests 506 taste/smell sensory domains may be responsible for similar patterns of food fussiness 507 that have been evidenced in children with neurodevelopmental disorders. It is clinically 508 important for future research to better understand how interventions, which take into 509 consideration taste/smell sensitivity impairments, may prevent or reduce food 510 fussiness.

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515

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519

# 520 7. References

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