#### **BRIEF REPORT**



# Brief Report: Odour Awareness in Young Children with Autism Spectrum Disorders

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#### **Abstract**

The elucidation of odour awareness in children with autism spectrum disorders (ASD) is important. We compared the odour awareness of young children with ASD with those of typical development (TD) children using the Children's Olfactory Behavior in Everyday Life (COBEL) questionnaire, which is a self-report measure that mainly assesses odour awareness. Forty-five young boys (aged 5-6 years), including 20 children with ASD and 25 TD children, participated in this study. The total COBEL score of the young children with ASD was lower than that of the TD children (p<0.01). Moreover, the total COBEL score was significantly correlated with the total VABS II score (p<0.05). Our results improve understanding of the odour awareness in children with ASD.

Keywords Autism spectrum disorders · Olfactory · Odour awareness · Self-report · VABS II

#### Introduction

Numerous studies have shown that compared to children with typical development (TD), children with autism spectrum disorders (ASD) experience increased sensory symptoms (Leekam et al. 2007; Tomchek and Dunn 2007). The recently released DSM-5 criteria (APA 2013) include sensory issues, reflecting the increasing interest in these symptoms in ASD. Odours have been shown to play an important role in inducing emotional reactions, imitating the actions of others, and regulating social interactions (Parma et al. 2013; Pause 2012; Soudry et al. 2011). Olfactory abnormalities

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might therefore contribute to the high rates of food refusal and selectivity in children with ASD (Hubbard et al. 2014). In addition, atypical responsiveness to olfactory stimuli has been reported to be the strongest predictor of social impairment in children with ASD (Hilton et al. 2007; Lane et al. 2010). Therefore, elucidating the odour awareness in children with ASD is important.

In contrast to abnormalities in touch, vision, and hearing, olfactory abnormalities in children with ASD remain poorly understood despite their importance. Experimental studies investigating olfactory abnormalities in ASD are limited. For example, researchers have investigated the olfactory detection thresholds in individuals with ASD in a laboratory setting (Ashwin et al. 2014; Dudova et al. 2011; Kumazaki et al. 2016; Suzuki et al. 2003; Tavassoli and Baron-Cohen 2012). However, the results of these studies are inconsistent (i.e., several studies reported no differences in the olfactory detection threshold, while other studies reported an enhanced or impaired detection threshold in individuals with ASD compared to that in controls). Originally, these previous studies used different types of odorants in olfactory detection threshold tests. While diverse (more than 400,000) odorants exist worldwide, in a laboratory setting, only a few odorants can be tested. Thus, children with ASD could be hyper-aware of certain odour stimuli and under-aware of other odour stimuli. Notably, to objectively measure sensory



abnormalities, investigations in experimental settings are essential. However, in the case of olfaction, because we are unconsciously affected by thousands of odours in daily life, the influence of the odours in daily life may not be directly observable or reproducible in laboratory settings.

Based on anecdotal reports and studies using sensory questionnaires, children with ASD may either not respond or only slightly respond to olfactory stimuli (high threshold) or, conversely, may be overwhelmed by stimuli (low threshold) (Kientz and Dunn 1997; Leekam et al. 2007; Rogers et al. 2003; Wiggins et al. 2009). These studies mainly focused on the other senses, such as vision and audition, or combined olfaction with taste; thus, information regarding pure olfactory function is limited. For example, the Sensory Profile questionnaire (Dunn 1994), which is a well-designed questionnaire consisting of 125 items, only includes four items related to pure olfaction and mainly focuses on sensorimotor defects. Moreover, these questionnaires were based on parental report. Notably, several limitations related to the exclusive use of a parental/proxy report should be considered. The fact that caregiver factors, such as stress, anxiety and depression, and Broader Autism Phenotype traits known to be prevalent among parents of individuals with ASD can significantly bias the reporting of a child's characteristics must be considered. Ben-Sasson et al. (2007) reported that parental report was not correlated with clinical observations of sensory issues. In addition, recognizing olfactory abnormalities is difficult, even for parents. From this perspective, assessments using self-reporting play an important role.

The Children's Olfactory Behavior in Everyday Life (COBEL) (Ferdenzi et al. 2008) was mainly developed to assess the awareness and uses of odours in real-life situations and to evaluate individual variation. This tool comprises 16 items prompting self-reports of awareness, active seeking and affective reactivity to odours of food (for instance, whether children try to guess what they will eat for dinner based on cooking smells), society (for instance, whether children realize that people have a natural odour), and environment (for instance, whether children seek out smells when they are feeling sad). Among these items, odours of society are of great interest because they include body odour, which plays an important role in imitation, especially for children with ASD (Parma et al. 2013, 2014). To the best of our knowledge, the COBEL is the only self-report tool used to assess olfaction in young children. The COBEL reduces task complexity and length, and by stimulating the children's attention and motivation (through the use of familiar vocabulary, a limited number of items, and variable response modes), users can easily obtain answers from even young children.

Age and IQ may be potential moderators of olfaction (Larsson et al. 2017). Chronological ageing plays an important role in olfactory studies investigating ASD

(Dudova and Hrdlicka 2013). Since the olfactory system has been suggested to develop differently in individuals with ASD than in controls (Brewer et al. 2008; May et al. 2011), age differences may explain the discrepant results. These confounding factors should be minimized in studies involving younger subjects within a narrow age range. A study involving younger children may help distinguish the primary features of the disorder from secondary effects associated with compensatory efforts and other factors. We must also consider the potential presence of sex differences in olfaction in children with ASD (Kumazaki et al. 2015). In this study, we included young boys aged 5–6, which is the minimum age at which children have the language mastery required to report details of their perceptions and affective reactions.

Few studies have investigated the relationship between odour awareness and adaptive behaviour in the daily lives of young children with ASD. To our knowledge, this is the first study to examine olfaction and its impact on daily life situations in young children with ASD using a self-report questionnaire. We predicted that our results could reflect a primary difference in olfactory features and the relationship between odour awareness and their effect on daily life in young children with ASD.

#### **Methods**

#### **Participants**

Participants were recruited from our institute well known in Japan for specializing in developmental disorders and related conditions. The participants were children with ASD and typical development. After a complete explanation of the study, all participants provided written, informed consent. All participants and their guardians agreed to participate in the study. The inclusion criteria for the participants were as follows: (1) male, (2) age 5–6 years, and (3) acquisition score of the The Kaufman Assessment Battery for Children (K-ABC) (Kaufman and Kaufman 1983) achievement score  $\geq$  70. The K-ABC was employed to estimate the intelligence levels of the children. The children with ASD were diagnosed using the Autism Diagnostic Observational Schedule-Generic (ADOS-G) (Lord et al. 2000), the Diagnostic Interview for Social and Communication Disorders (DISCO) (Wing et al. 2002), and the DSM-5 criteria upon entering this study. The children in the ASD group were included in this study if they fulfilled the criteria for a diagnosis of childhood autism, atypical autism or Asperger's syndrome with DISCO or met the ADOS criteria for autism spectrum disorders. TD participants had no first-degree relatives with a diagnosis of ASD.



# **COBEL Questionnaire**

The COBEL (Ferdenzi et al. 2008) was mainly developed to assess odour awareness and uses of odours in real-life situations and to evaluate individual variation. This tool comprises 16 items prompting self-reports of awareness of, active seeking of and affective reactivity to the odours of food, people and the environment. In the original paper (Ferdenzi et al. 2008), participants were 6-10 years old. However, Martinec Novakova et al. (2018) used the COBEL questionnaire with children 4.33-6.92 years old and Martinec Nováková and Vojtušová Mrzílková (2016) used the COBEL questionnaire with children 5.25-6.75 years old. In our preliminary experiment, we confirmed that 5-year-old children could respond to the COBEL interview. The Japanese version of the COBEL was prepared using a forward/ backward translation method. The comprehensibility of the translated version was confirmed by parents and children in a pilot study. Please refer to the supplementary materials for the content of the COBEL. The COBEL data of all participants were obtained during interviews with the parents.

### **Questionnaires and Interview Content**

The quantitative autistic traits of the enrolled children were assessed by the parents using the Japanese version of the Social Responsiveness Scale, Second Edition (SRS-2) (Kamio et al. 2013). Higher scores on the SRS-2 indicate a higher degree of social impairment. The raw scores of the SRS-2 were converted to T-scores (with a mean of 50 and a standard deviation of 10). The SRS-2 was completed by the parents of all participants. The parents of the children in the TD group also completed the Social Communication Questionnaire (SCQ; Rutter et al. 2010) to screen for clinically significant ASD symptoms in the TD group. Furthermore, to exclude psychiatric diagnoses, the Mini-International Neuropsychiatric Interview for Children and Adolescents (MINI Kids) (Otsubo et al. 2005; Sheehan et al. 1998) was administered.

The Vineland Adaptive Behavior Scale II (VABS II) is a semi-structured interview administered to caregivers to assess adaptive behaviour (Sparrow et al. 2005). Adaptive behaviour is defined by this instrument as the development and application of abilities required for the attainment of personal independence and social competence. The VABS II consists of five domains, i.e., daily living, communication, socialization, motor skills and maladaptive behaviour. The items in each domain are scored from zero to two, with lower scores indicating skills/behaviours that are occasionally or never performed. The standard scores and an adaptive behaviour composite score can be calculated to reflect the overall ability of the participants to live independently. The VABS II has been shown to have adequate internal

consistency and reliability, good test—retest reliability and excellent inter-rater reliability, with coefficients ranging between the .80 s and high .90 s (Sparrow et al. 2005). The VABS II was also demonstrated to have good construct, content and criterion-related validity (Sparrow et al. 2005). The VABS II data of all participants were obtained by interviews with the parents.

### **Data Analysis**

We performed the statistical analyses using SPSS version 24.0 (IBM, Armonk, NY, USA). The descriptive statistics of the sample were analysed. The differences between the groups in age, K-ABC achievement score, and SRS were analysed by performing an independent samples t-test. The differences in the total scores and in each COBEL-item score and in the total scores and subscale scores of the VABS II were analysed by performing a Mann–Whitney U test comparing the TD and ASD children. We performed a Spearman's rank correlation analysis to explore the relationships between the COBEL score and age, SRS score, K-ABC achievement score, and VABS II total and subscale scores. An alpha level of 0.05 was employed for these analyses.

#### Results

## **Demographic Data**

The ASD group included 20 participants with a mean age of  $70.80 \pm 6.44$  months. The TD group included 25 participants with a mean age of  $67.72 \pm 5.86$  months. No significant differences in mean age were observed between the groups (t=-1.678; df=43; p=0.10). Expectedly, significant differences were observed in the SRS-2 (t=-5.463; df=43; p<0.01) and K-ABC achievement (t=2.586; df=43; p=0.01) scores between children with ASD and TD children. The participants' details are presented in Table 1.

#### **Main Data**

Regarding the VABS II, significant differences were observed in the total (z=-3.876; p<0.01), daily living (z=-2.532; p=0.01), communication (z=-3.951; p<0.01), social (z=-3.487; p<0.01), motor (z=-3.480; p<0.01), and maladaptive behaviour (z=4.132; p<0.01) scores between the children with ASD and TD children. The details are presented in Table 2.

All participants completed the COBEL questionnaires, which require approximately 10 min to complete. Significant differences were observed in the total score of the COBEL questionnaires (The ASD group:  $3.03 \pm 0.59$ , The TD group:  $4.96 \pm 0.47$ ; z = -3.050; p < 0.01). In order to characterize



**Table 1** Participant characteristics

	Age (months)	SRS-2	SCQ	K-ABC achievement score
ASD (n=20) mean (SD)	70.80 (6.44)	49.08 (8.48)		91.95 (16.14)
TD $(n=25)$ mean $(SD)$	67.72 (5.86)	66.55 (12.90)	2.92 (1.89)	103.76 (14.46)

SD standard deviation, SRS-2 social responsiveness scale, SCQ social communication questionnaire lifetime total score, K-ABC the Kaufman assessment battery for children

**Table 2** Participants' performance on the VABS II

Item	ASD (n=20) mean (SEM)	TD (n=25) mean (SEM)	Mann-Whitney U test	
			$\overline{U}$	p
Total	83.70 (16.95)	106.24 (13.75)	80.5	< 0.01**
Daily living	90.45 (14.76)	101.00 (12.84)	139.5	0.01*
Communication	84.25 (14.53)	103.68 (12.07)	77.5	< 0.01**
Social	85.70 (25.62)	112.88 (18.50)	97.5	< 0.01**
Motor	80.15 (15.62)	97.44 (12.06)	98.0	< 0.01**
Maladaptive behaviour	18.80 (2.02)	15.76 (2.02)	430.5	< 0.01**

*VABS II* vineland adaptive behavior scales II, *SEM* standard error of the mean \*p < 0.05, \*\*p < 0.01

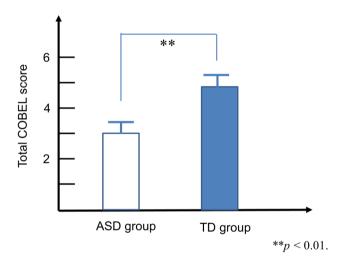


Fig. 1 Significant differences in the total COBEL score between the children with ASD and TD children

children's olfactory behavior in detail, each COBEL item score was examined, as was performed in the previous study using the COBEL (Ferdenzi et al. 2008). Significant differences between ASD and TD groups were found in item 8: smelling school tools (The ASD group:  $0.08 \pm 0.05$ , The TD group:  $0.28 \pm 0.07$ ; z = -2.561; p = 0.01), and item 12: people's natural odour (The ASD group:  $0.05 \pm 0.03$ , The TD group:  $0.34 \pm 0.08$ ; z = -2.803; p < 0.01) scores between the children with ASD and TD children. The details are presented in Fig. 1 and Table 3.

The Spearman's rank correlation analysis revealed significant negative correlations between the total COBEL score and the total SRS-2 total score (r = 0.42, p < 0.01) and

significant positive correlations between the total COBEL total score and the total (r = 0.32, p = 0.04), daily living (r = 0.30, p < 0.05), and motor (r = 0.30, p < 0.05) scores on the VABS II. The details are presented in Table 4.

#### Discussion

The aim of this study was to examine odour awareness using a self-report questionnaire (COBEL) that was mainly developed to assess the awareness and uses of odours in real-life situations. All participants completed the questionnaire, suggesting that this tool was a feasible measure of odour awareness, even in young children with ASD. In previous studies (Ferdenzi et al. 2008), the total COBEL score of 6-year-old boys has been found to be approximately 5; thus, the results of the TD participants in our study are similar to those reported in previous studies. Using this self-report questionnaire, we found that children with ASD displayed lower total COBEL scores than TD children. Ben-Sasson et al. (2007) found that children with ASD showed a lower awareness of sensations than typically developing children. Our results are consistent with these findings, the children with ASD in this study had a lower awareness of sensation. In addition, according to our data, lower COBEL scores may be related to autistic traits and lower levels of adaptive behaviour in daily living.

Odour awareness is strongly associated with better olfactory abilities and higher reactivity to smells (Buron et al. 2015). Odour awareness is also positively associated with the impact of odours on emotions, cognition and behaviour (Burón et al. 2011; Buron et al. 2013). Thus, investigating



**Table 3** COBEL score of the children in the ASD and TD groups

Item	ASD $(n=20)$ mean	TD $(n=25)$ mean	Mann-Whitney U test	
	(SEM)	(SEM)	$\overline{U}$	p
Odour of disliked food	0.08 (0.05)	0.14 (0.07)	234.0	0.54
2. Response to unknown food	0.30 (0.08)	0.44 (0.07)	191.5	0.15
3. Senses in nature	0.25 (0.09)	0.26 (0.07)	236.0	0.71
4. Yesterday's odours	0.25 (0.09)	0.34 (0.09)	223.0	0.48
5. Odours sought when sad	0.10 (0.05)	0.18 (0.06)	226.0	0.46
6. Treasured odorous objects	0.13 (0.06)	0.24 (0.07)	208.5	0.23
7. Outside odours	0.20 (0.08)	0.36 (0.08)	189.5	0.12
8. Smelling school tools	0.08 (0.05)	0.28 (0.07)	159.0	0.01*
9. Odours in cars	0.30 (0.08)	0.34 (0.08)	238.0	0.76
10. Odour of bathroom objects	0.25 (0.77)	0.32 (0.08)	231.0	0.62
11. Family odours	0.30 (0.09)	0.34 (0.08)	234.0	0.68
12. People's natural odour	0.05 (0.03)	0.34 (0.08)	150.0	< 0.01**
13. Smelling clothes	0.15 (0.07)	0.32 (0.08)	191.0	0.11
14. Smelling self-odour	0.13 (0.06)	0.22 (0.06)	203.0	0.18
15. Tobacco smell	0.24 (0.09)	0.50 (0.09)	162.5	0.05
16. Guessing food odour	0.18 (0.07)	0.34 (0.06)	176.5	0.06

SEM standard error of the mean

Table 4 Correlations COBEL total score and age, SRS-2 total score, K-ABC achievement score and total and subscale scores on the VABS II

Item	COBEL total score
Age (month)	-0.23
SRS-2 total	-0.42**
K-ABC achievement score	0.17
VABS	
Total	0.32*
Daily living	0.30*
Communication	0.29
Social	0.23
Motor	0.30*
Maladaptive behaviour	-0.27

SRS-2 social responsiveness scale, K-ABC the Kaufman assessment battery for children, VABS II Vineland adaptive behavior scales II \*p < 0.05, \*\*p < 0.01

odour awareness in ASD may provide a better understanding of this clinical phenomena.

In this study, particularly on item 12 of the COBEL, i.e., people's natural odour, young children with ASD displayed lower odour awareness than the TD children. Thus, young children with ASD and TD children have different perceptions of body odours. Previous studies have suggested that body odours have a large impact on social behaviour in children with ASD (Parma et al. 2013, 2014). One plausible theory might be that different odour awareness results in differences in social behaviour. However, the influence of

odours on behaviour is, in fact, greater when the odours are subliminal than when they are consciously perceived (Cecchetto et al. 2017; Li et al. 2007). Endevelt-Shapira et al. (2018) revealed that TD and ASD participants dissociated their responses to subliminal presentation of body odours. Therefore, future studies investigating the relationship between both subliminal and supraliminal odours and social behaviour in children with ASD and TD children are needed.

Because sensory abnormalities, including olfaction abnormalities, are currently used as a diagnostic tool for identifying ASD early in life and because of the profound impact of such abnormalities on those affected, developing a more comprehensive understanding of the emergence of olfactory abnormalities early in development is crucial. In addition, sensory abnormalities, including olfaction abnormalities, may represent a key physiological factor underlying the social impairments associated with ASD. The early perceptual capacities of individuals with ASD have been proposed to trigger a cascade of developmental deficits that contribute to the poor social skills observed at older ages (Leekam et al. 2007). Thus, assessing the COBEL scores at an early age may provide important information.

Several limitations in our study should be acknowledged. First, the number of participants was relatively small. In addition, all participants were boys. Although the results of the TD group are similar to the results reported in previous studies (Ferdenzi et al. 2008), future studies involving larger sample sizes and female participants are needed to provide more meaningful data. Second, in this study, the cognitive skills of the participants were in the average range. All participants



p < 0.05, p < 0.01

in this study completed the questionnaire. Participants with a below average IQ may not have been able to complete the questionnaire. Therefore, future studies involving participants with a broader range of cognitive abilities are necessary to obtain a richer understanding of odour awareness in young children. Thirdly, we used only one questionnaire for sensory (olfactory) functions in this study. Future studies are needed to compare the results of the COBEL questionnaire with those of some other well-established questionnaire for sensory (olfactory) function assessment or with those of some odour test for children.

In summary, in this study, the COBEL score, which mainly assesses the awareness and uses of odours in real-life situations, was lower in children with ASD than in TD children. Furthermore, the COBEL score is related to adaptive behaviour in daily life situations. Considering the role played by odours in daily life, future studies investigating odour awareness in children with ASD using this questionnaire may allow for the elucidation of the mechanism of ASD.

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Author contributions HK designed the study, conducted the experiment, performed the statistical analyses, analysed and interpreted the data, and drafted the manuscript. MO, YY, TI, CH, DS, RI, ST, AK, YM and MK conceived the study, participated in its design, assisted with the data collection and scoring of the behavioural measures, analysed and interpreted the data, participated in the drafting of the manuscript and critically revised the manuscript for important intellectual content. MK was involved in the final approval of the version to be published. All authors read and approved the final manuscript.

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# **Compliance with Ethical Standards**

Conflict of interest All the authors declares that they have no conflict of interest.

**Ethical Approval** All procedures involving human participants were conducted in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

**Informed Consent** Participants were recruited from Kanazawa University. After a complete explanation of the study, all the participants and their parents provided written, informed consent. All participants and their parents agreed to participate in the study.

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