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Brief Report

Sensory processing profiles and autistic symptoms as predictive factors in autism spectrum disorder and Williams syndrome

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Abstract

Background Unusual sensory responses were included in the diagnostic criteria for autism spectrum disorder (ASD), yet they are also common among individuals with other neurodevelopmental disorders, including Williams syndrome (WS). Cross-syndrome comparisons of sensory atypicalities and the evaluation of their syndrome specificity however have rarely been undertaken. We aimed to (1) examine and compare the sensory profiles in ASD and WS groups and (2) investigate whether autistic symptoms, including sensory processing scores, can predict a group membership.

Methods Parents of 26 children with ASD and intellectual disability, 30 parents of children with ASD (no intellectual disability) and 26 with WS aged between 4 and 16 years were recruited. Parents completed the Sensory Profile to provide information about their children's sensory experiences and the Social Responsiveness Scale – Second Edition (SRS-2) to assess the degree of social impairment in their children.

Results No significant differences were found in sensory processing scores between the three groups. Binary logistic regression analyses were undertaken with sensory quadrants and SRS-2 total score as factors. Models significantly predicted group membership, with Low Registration, Sensory Sensitivity and SRS-2 total score being significant predictors.

Conclusions The findings suggest that high rates of sensory atypicalities are a common neurodevelopmental characteristic that do not reliably distinguish between WS and ASD groups. Low Registration and Sensory Sensitivity-related behaviours might, however, be more specific to ASD. Further work is needed to explore what behaviours within sensory profiles can discriminate between neurodevelopmental disorders and should be included in diagnostic classifications.

Keywords autism spectrum disorder, sensory processing, Sensory Profile, Social Responsiveness Scale, socio-communicative behaviours, Williams syndrome

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Introduction

Alongside impairments in social communication and the presence of restricted and repetitive interests and behaviours, atypical sensory processing is a diagnostic feature of autism spectrum disorder (ASD; Diagnostic and Statistical Manual of Mental Disorder-5, American Psychiatric Association 2013). It has been associated not only with other core features of ASD but also with other behavioural and emotional characteristics of the disorder, such as the presence of enhanced attention to detail or heightened anxiety (Green & Ben-Sasson 2010; Green, Ben-Sasson, Soto, & Carter 2012; Lane, Reynolds, & Dumenci 2012; for a review, see Glod, Riby, Honey, & Rodgers 2015).

Williams syndrome (WS) is a rare neurodevelopmental disorder caused by the microdeletion of approximately 17–28 genes on chromosome 7q11.23 (Donnai & Karmiloff-Smith 2000; Osborne 2006). WS is characterised by mild to moderate intellectual disability (ID; Searcy *et al.* 2004), distinctive facial features (Donnai & Karmiloff-Smith 2000) and cardiovascular difficulties (Morris 2006). The disorder is also associated with unusual cognitive profile and personality features such as particular problems with spatial tasks, but stronger language skills and hypersociability (Jones *et al.* 2000; John & Mervis 2010).

High levels of sensory sensitivity in WS compared with children with other developmental disorders, including ASD, have been reported (Klein-Tasman & Mervis 2003) and demonstrated in maladaptive physical and/or emotional reactions to everyday stimuli; 85–95% of individuals with WS have been reported as frequently showing sensitivity to the sounds of machines, fireworks and bursting balloons (Donnai & Karmiloff-Smith 2000). More recently, WS has also been linked to a greater range of sensory processing atypicalities (Rodgers, Riby, Janes, Connolly, & McConachie 2012; Riby, Janes, & Rodgers 2013).

In addition to the growing evidence of the pervasiveness of sensory processing atypicalities across neurodevelopmental disorders, there is mixed evidence in relation to the syndrome specificity of socio-communicative abnormalities in ASD, as assessed by the Social Responsiveness Scale (SRS; Constantino & Gruber 2005). There is evidence that socio-communicative abnormalities are common in children with WS (Klein-Tasman, Mervis, Lord, & Phillips 2007) and these atypicalities are very similar to the difficulties observed in children with ASD (Klein-Tasman, Li-Barber, & Magargee 2011). Indeed, a co-morbid autism diagnosis has been reported in several WS cases (Gillberg & Rasmussen 1994; Herguner & Motavalli Mukaddes 2006). Also, Riby *et al.* (2014) showed that 58% of individuals with WS (reported by parent-completed SRS) had severe deficits of reciprocal social interaction.

There is emerging evidence that some ASD symptoms, including sensory processing atypicalities and social communication impairments, are also common in WS. A cross-syndrome comparison of the social communication features and sensory profiles of the two disorders is lacking. It is needed to establish trans-diagnostically unique discriminative sensory difficulties and social communication features and shared characteristics of these conditions in order to contribute to both developmental theory and disorder-specific or shared intervention programmes.

The aims of this study were (1) to examine and compare the sensory and social responsiveness profiles in three groups of children and adolescents with a neurodevelopmental disorder, those with a diagnosis of ASD with ID (ASD + ID), those with ASD without additional ID and those with WS and (2) to investigate whether social communication features and sensory processing scores can predict group membership. The groups were chosen to facilitate cross-syndrome comparisons of social communication and sensory processing across both ASD and WS with consideration of different ability level in both disorders. Distinguishing two ASD groups (ASD and ASD + ID) allows us to examine and establish unique discriminative features of the disorders regardless of the level of intellectual ability.

Methods

Participants

Parents of children with ASD or WS between 4 and 16 years of age were recruited to the study via ASD-UK (www.ASD-UK.com), a representative, large national UK family research database of children with ASD (Warnell *et al.* 2015); 'Contact', a

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national UK charity for families with disabled children; the Williams Syndrome Foundation (WSF; UK charity 281014); and local mainstream and special schools. The initial dataset consisted of 35 parents of children with WS and 74 parents of children with ASD. Children had no diagnosed sensory impairments. Due to a large amount of missing data, the final samples consisted of 26 children with WS and 56 children with ASD, of whom 26 had additional ID.

The WS children had their diagnosis confirmed genetically with fluorescent in situ hybridisation testing. Diagnoses of ASD were based on a multidisciplinary team assessment following the guidelines of the UK National Autism Plan for Children (Le Couteur 2003), as stated by the parents. Within the ASD sample, two subgroups were distinguished – those with ASD without additional diagnosis of ID and children with ASD with co-morbid ID. ID status was either assigned in agreement with a parent report based on previous formal diagnosis given by a clinician (for 43 children, data were available, and 22 ASD children were classified as having additional ID) or examined in a direct assessment using both Raven's Coloured Progressive Matrices (Raven, Raven, & Court 1998) and British Picture Vocabulary Scale – Third Edition (Dunn *et al.* 2009). Those who obtained standard scores below 70 on both measures were identified as having additional ID (Table 1).

Measures

Sensory Profile (Caregiver Questionnaire) (SP; Dunn 1999) measures children's sensory processing abilities. The questionnaire consists of 125 items, rated on a 5-point Likert scale, ranging from almost never to almost always. Children can be classified as fitting into one of the four sensory processing quadrants: Sensation Seeking (actively looking for ways of increasing sensory input in everyday situations), Sensation Avoiding (actively limiting or avoiding the number of sensory stimuli), Sensory Sensitivity (readily responding to sensory events that are not detected or noticeable by other people) and Low Registration (not responding to some of usual sensory events that other people notice easily and respond to). The items include 'Has trouble completing tasks when the radio is on', 'Prefers to be in the dark' or 'Touches people and objects'. Lower scores on the SP indicate greater level of difficulties. Cronbach's alpha for the current cohorts is presented in Table 2.

Social Responsiveness Scale – Second Edition (SRS-2; Constantino & Gruber 2012) is a 65-item parent-report rating scale of autistic traits in 4- to 18-year-olds. The items cover a range of unusual interpersonal behaviours, communication or repetitive/stereotyped behaviours and are rated on a 4-point Likert scale, ranging from not true to almost always true. The SRS-2 provides scores in five subscales including Social Awareness, Social Cognition, Social Communication, Social Motivation and Restricted Interests and Repetitive Behaviour (RRB), and the Total score. Higher scores indicate greater impairment. Cronbach's alpha for the SRS-2 for the current cohorts is presented in Table 2.

Data analysis

To determine whether there were significant differences between the means of the three groups and to investigate whether autistic symptoms and sensory quadrant scores could predict group membership one-way analysis of variance, chi-square and regression analyses were subsequently undertaken on the complete dataset using SPSS

Table 1 Descriptive statistics of participant characteristics

	WS (*n* = 26)	**ASD + ID (*n* = 26)**	**ASD (*n* = 30)**
Gender: male	13	24	21
Age in months: mean (SD)	96.77 (35.71)	108.85 (39.36)	90.10 (27.11)
Age in months: range	53–181	54–184	50–161

ASD, autism spectrum disorder; ID, intellectual disability; SD, standard deviation; WS, Williams syndrome.

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Table 2 Cronbach's alpha values for the Social Responsiveness Scale – Second Edition and Sensory Profile subscales

Cronbach's alpha	WS (*n* = 26)	ASD + ID (*n* = 26)	ASD (*n* = 30)
SRS-2			
Social Awareness	0.632	0.598	0.669
Social Cognition	0.806	0.697	0.728
Social Communication	0.848	0.849	0.866
Social Motivation	0.773	0.873	0.833
RRB	0.828	0.800	0.843
SP quadrant			
Low Registration	0.915	0.897	0.938
Sensation Seeking	0.819	0.920	0.851
Sensory Sensitivity	0.888	0.876	0.782
Sensation Avoiding	0.897	0.876	0.871

ASD, autism spectrum disorder; ID, intellectual disability; RRB, Restricted Interests and Repetitive Behaviour; SP, Sensory Profile; SRS-2, Social Responsiveness Scale – Second Edition; WS, Williams syndrome.

version 22. Adaptions for multiple comparisons were dealt with by applying Bonferroni corrections.

Results

The groups did not differ on the mean age ($F_{2,79}$ = 2.14, P = 0.125); however, they did differ on gender ($\chi^2_{(2)}$ = 10.14, P = .005) with the ASD + ID group having significantly more male participants than the WS group (24 to 13, P = 0.002). Descriptive statistics for the participants on the outcome variables are presented in Table 3, and the autism traits severity categories are presented in Table 4.

The groups were significantly different on most of the SRS-2 subscales, but Social Cognition ($F_{2,79}$ = 1.35, P = 0.265) and RRB ($F_{2,79}$ = 2.06, P = 0.134). For the SRS-2 total score, the WS group differed from both ASD groups, with significantly lower scores. The same pattern was found for the SRS-2 subscales, with the WS sample having significantly lower scores than both ASD groups. For the SP, the groups were significantly different on the Low Registration ($F_{2,79}$ = 3.73, P = 0.028) only (Bonferroni test: WS and ASD, P = 0.049, WS and ASD + ID, P = 0.074), while the differences in mean scores on Sensation Seeking ($F_{2,79}$ = .69, P = 0.505), Sensory Sensitivity ($F_{2,79}$ = .65, P = 0.524) and Sensation Avoiding ($F_{2,79}$ = .27, P = 0.763) were not significant.

As post hoc analysis indicated, for Low Registration only the WS and the ASD groups differed significantly (P = 0.042) with the WS group having lower scores than the ASD group.

Multinomial logistic regression was undertaken with sensory quadrants and SRS-2 subscales as covariates. The model was significant ($\chi^2_{(18)}$ = 91.62, P < .001). Low Registration ($\chi^2_{(2)}$ = 23.57, P < 0.001), Sensation Avoiding ($\chi^2_{(2)}$ = 8.82, P = 0.012), Social Communication ($\chi^2_{(2)}$ = 8.75, P = 0.013), Social Awareness ($\chi^2_{(2)}$ = 8.00, P = 0.021) and Social Cognition ($\chi^2_{(2)}$ = 8.26, P = 0.016) had a significant

Table 3 Mean (standard deviation) scores on outcome variables

Variable	WS (*n* = 26)	ASD + ID (*n* = 26)	ASD (*n* = 30)
SRS-2			
Total raw score	87.85 (28.72)	118.81 (29.76)	111.80 (30.38)
Social Awareness	11.54 (3.74)	15.85 (3.81)	14.43 (4.01)
Social Cognition	20.00 (5.79)	22.54 (5.99)	20.60 (5.76)
Social Communication	26.42 (9.69)	40.00 (10.92)	37.63 (10.09)
Social Motivation	10.04 (5.90)	18.58 (6.83)	17.17 (7.63)
RRB	19.85 (7.50)	23.92 (7.39)	21.97 (6.89)
SP quadrant			
Low Registration	43.85 (13.51)	52.23 (13.89)	52.53 (12.29)
Sensation Seeking	87.62 (17.07)	82.08 (16.07)	82.97 (21.14)
Sensory Sensitivity	61.65 (14.56)	65.58 (11.14)	65.23 (15.27)
Sensation Avoiding	90.65 (17.89)	92.12 (16.21)	88.77 (17.05)

Lower scores indicate better performance on the SRS and greater level of difficulties on the SP. ASD, autism spectrum disorder; ID, intellectual disability; RRB, Restricted Interests and Repetitive Behaviour; SP, Sensory Profile; SRS-2, Social Responsiveness Scale – Second Edition; WS, Williams syndrome.

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Table 4 Severity group profiles on both the Social Responsiveness Scale – Second Edition and Sensory Profile with number of individuals (percentage) falling into each category

Variable	WS (*n* = 26)	ASD + ID (*n* = 26)	ASD (*n* = 30)
SRS-2 Total score			
Typical	3 (11.5)	–	–
Mild	3 (11.3)	2 (7.7)	2 (6.7)
Moderate	9 (34.6)	5 (19.2)	7 (23.3)
Severe	11 (42.3)	19 (73.1)	21 (70.0)
Low Registration			
Similar to others	3 (11.5)	5 (19.2)	6 (20.0)
More than others	3 (11.5)	4 (15.4)	5 (16.7)
Much more than others	20 (79.6)	17 (65.4)	19 (63.3)
Sensation Seeking			
Less than others	–	–	1 (3.3)
Similar to others	4 (15.4)	–	3 (10.0)
More than others	9 (34.6)	10 (38.5)	6 (20.0)
Much more than others	13 (50.0)	16 (61.5)	20 (66.7)
Sensory Sensitivity			
Similar to others	2 (7.7)	3 (11.5)	5 (16.7)
More than others	7 (26.9)	6 (23.1)	7 (23.3)
Much more than others	17 (65.4)	17 (65.4)	18 (60.0)
Sensation Avoiding			
Similar to others	3 (11.5)	2 (7.7)	2 (6.7)
More than others	4 (15.4)	5 (19.2)	3 (10.0)
Much more than others	19 (73.1)	19 (73.1)	25 (83.3)

ASD, autism spectrum disorder; ID, intellectual disability; SRS-2, Social Responsiveness Scale – Second Edition; WS, Williams syndrome.

main effect on diagnostic group. Non-significant main effects were found for Sensory Seeking ($\chi^2_{(2)} = 0.40$, $P = 0.82$), Sensory Sensitivity ($\chi^2_{(2)} = 4.18$, $P = 0.123$), RRB ($\chi^2_{(2)} = 1.12$, $P = 0.572$) and Social Motivation ($\chi^2_{(2)} = 1.87$, $P = 0.392$). Given the difference in gender distribution across the groups, gender was added to the model; however, the main effect of gender was non-significant ($\chi^2_{(2)} = 3.86$, $P = 0.144$).

As indicated by parameter estimates with the WS group as a comparison, Low Registration ($\beta = 0.43$, Wald $\chi^2_{(1)} = 5.33$, $P = 0.021$), Social Cognition ($\beta = -0.54$, Wald $\chi^2_{(1)} = 4.71$, $P = 0.03$) and Social Communication ($\beta = -0.56$, Wald $\chi^2_{(1)} = 4.71$, $P = 0.041$) significantly predicted whether a participant had WS or ASD. The odds ratio indicated that as Low Registration and Social Communication increased in unit and Social Cognition decreased in unit, a participant was more likely to be diagnosed with ASD rather than with WS. Only Low Registration significantly predicted whether an individual had WS or ASD + ID ($\beta = 0.42$, Wald $\chi^2_{(1)} = 5.10$, $P = 0.024$), with the odds ratio showing that as Low Registration increased, it was more likely for a child to be diagnosed with ASD + ID rather than WS. The summary of the results is presented in Table 5.

Additionally, when the ASD + ID group was placed in the model as the comparison group, the only variable that significantly predicted whether a participant had a diagnosis of ASD + ID or ASD was Sensation Avoiding ($\beta = 0.04$, Wald $\chi^2_{(1)} = 4.25$, $P = 0.039$) with the odds ratio showing that as Sensation Avoiding increased, it was more likely for a child to be diagnosed with ASD + ID rather than ASD.

To control for a possible effect of age, additional multinomial regression was undertaken with child age entered as a covariate. The model was significant ($\chi^2_{(20)} = 95.77$, $P < 0.001$); however, the main effect of age was non-significant ($\chi^2_{(2)} = 4.15$, $P = 0.125$). The summary of the results is presented in Table S1.

Discussion

The aim of this study was to examine and compare the sensory and social responsiveness profiles in three groups of children and adolescents with a neurodevelopmental disorder and investigate whether autistic symptoms and sensory processing scores can predict group membership.

Parent reports of children's sensory processing and social communication features indicated that only Low Registration from the SP and Social Cognition and Social Communication subscales from the SRS-2 were significant predictors of whether a child had WS or ASD. While, only Low Registration scores significantly predicted whether an individual had WS or ASD + ID, suggesting that these groups were similar in their sensory and social responsiveness profiles.

The WS group obtained significantly lower (still within atypical range) scores on the SRS-2 than both ASD groups. Although some authors report that SRS scores can distinguish children with pervasive

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Table 5 Multinomial logistic regression

	β (SE)	95% CI for odds ratio			*P* value
		Lower	Odds ratio	Upper	
WS vs. ASD					
Intercept	−29.17 (20.48)				0.154
Low Registration	0.43 (19)*	1.07	1.53	2.21	0.021
Sensation Seeking	−0.03 (0.06)	0.86	0.97	1.09	0.627
Sensory Sensitivity	0.18 (0.12)	0.96	1.20	1.51	0.112
Sensation Avoiding	−0.18 (0.10)	0.69	0.84	1.02	0.071
Social Awareness	0.90 (0.58)	0.80	2.46	7.62	0.118
Social Cognition	−0.54 (0.25)*	0.36	0.58	0.95	0.030
Social Communication	0.56 (0.27)*	1.02	1.74	2.97	0.041
Social Motivation	0.17 (0.17)	0.86	1.19	1.65	0.302
RRB	−0.21 (0.22)	0.53	0.81	1.25	0.341
WS vs. ASD + ID					
Intercept	−41.47 (20.82)*				0.046
Low Registration	0.42 (19)*	1.06	1.52	2.20	0.024
Sensation Seeking	−0.02 (0.06)	0.87	0.98	1.11	0.751
Sensory Sensitivity	0.17 (0.12)	0.94	1.19	1.49	0.146
Sensation Avoiding	−0.09 (0.10)	0.75	0.91	1.11	0.345
Social Awareness	1.06 (0.58)	0.92	2.88	8.97	0.069
Social Cognition	−0.42 (0.26)	0.40	0.66	1.09	0.101
Social Communication	0.52 (0.27)	0.99	1.68	2.86	0.057
Social Motivation	0.21 (0.17)	0.88	1.24	1.73	0.216
RRB	−0.17 (0.22)	0.55	0.85	1.30	0.442

*$P < 0.05$.
R^2 = 0.67 (Cox and Snell), 0.76 (Nagelkerke). ASD, autism spectrum disorder; CI, confidence interval; ID, intellectual disability; RRB, Restricted Interests and Repetitive Behaviour; SE, standard error; WS, Williams syndrome.

developmental disorders from those with other disorders (Constantino, Przybeck, Friesen, & Todd 2000; Constantino & Todd 2000, 2003), others report commonality of socio-communicative abnormalities in children with WS (Klein-Tasman *et al.* 2007) and similarity to difficulties present in children with ASD (Klein-Tasman *et al.* 2011; for the review, see Vivanti, Hamner, & Lee 2018). The results indicate that there was a relationship between difficulties in Social Communication and Social Cognition (as assessed by the SRS-2) and a diagnosis of either ASD or WS; however, the relationships differ in their direction. We found that an increase in Social Communication difficulties and a decrease in Social Cognition scores were associated with ASD rather than WS. The Social Communication scale assesses reciprocity of social communication (e.g. ability to keep the flow of a conversation) (Bruni 2014), and this in turn relates to social communication and interaction, one of the two core diagnostic symptoms of autism (Frazier, Youngstrom, Kubu, *et al.* 2008; Snow, Lecavalier, & Houts 2009; Frazier *et al.* 2012). That supports the notion of the crucial role of the social communication behaviours in ASD. Social Cognition that assesses processing of social information (e.g. understanding meaning of social behaviours) (Bruni 2014) has been linked to the likelihood of diagnosis of WS rather than ASD, and previously, parents of children with WS also rated significantly higher (more problematic) Social Cognition than Social Communication (Klein-Tasman *et al.* 2011). It is worth noticing that Social Communication, after controlling for age, did not any longer significantly predicted group membership.

It is worth noting that neither Social Communication nor Social Cognition, nor any other SRS-2 subscales, significantly predicted either WS or ASD + ID diagnosis. This is theoretically and

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clinically important. ID is a characteristic of both of these groups, indicating that for those with an autism diagnosis, their social communication and social cognition difficulties (as measured by the SRS-2) are not distinguishable from those with WS. Further, more detailed research, which involves direct assessment of these skills, is needed to replicate these results and establish whether social responsiveness profiles can discriminate between the neurodevelopmental groups.

The findings of this study also suggest that sensory symptoms in children and adolescents with WS and ASD are very similar. Only Low Registration, associated with a high threshold for sensory experiences, passive responses to sensory events and limited detection of changes in sensory situations (Dunn 1997), predicted whether a child had WS or ASD. Decreased scores increased the likelihood for a child to be diagnosed with WS rather than ASD + ID, which is in contrast with previous findings (Baranek, David, Poe, Stone, & Watson 2006). What is clear is that heterogeneity is an important feature of both conditions and individual variability in sensory processing should be always considered with assumptions regarding the occurrence of particular sensory profiles or patterns in ASD or WS should be made with caution. Further research investigating sensory profiles in ASD and WS and examining cross-syndrome comparisons is needed to establish shared and unique discriminative sensory difficulties for each of these conditions in order to enhance our understanding of the gene–brain–behaviour relationships in neurodevelopmental disorders.

There are several notable limitations of the current study. First, although in the current study three groups of children with neurodevelopmental disorders were included, the sample sizes were relatively small. This is particularly important as 10 cases per predictor are recommended for logistic regression analysis, and thus, this study is underpowered (Tabachnick & Fidell 2019) Second, only parent reports were used in this investigation. It is important to note that the SRS-2 was designed solely for use with ASD and was not aimed to be discriminable. It is difficult to know whether parents are taking the same frame of reference when they respond to describe their children's behaviours. The questions focus on the presence rather than a nature of an atypicality, and therefore, more fine social behaviours present in the WS might not be reported (such as the atypical increased social motivation in WS; Lough, Rodgers, Janes, Little, & Riby 2016). Moreover, data from multiple raters and measures, including direct assessments of social behaviours and sensory processing, would provide better understanding of children's strengths and difficulties. Finally, only a limited number of predictors were entered into the model. However, there are possibly other features that can change the likelihood of WS or ASD diagnosis, for example, co-morbid behaviour problems that were found to influence autism symptoms (Mayes & Calhoun 2010; Hus, Bishop, Gotham, *et al.* 2013). Incorporating more symptoms could enhance the power of the model.

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Compliance with Ethical Standards

Ethical approval

All procedures performed in studies involving human participants were 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

Informed consent was obtained from all individual participants included in the study.

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Conflict of Interest

The authors report no real or potential conflicts of interest.

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Supporting Information

Additional Supporting Information may be found online in the supporting information tab for this article.

Table S1. Multinomial logistic regression including age as a covariate

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