Comparison of Gustatory Over-Responsivity Between Parent-Report and Clinician-Rated Assessment in a Pediatric Cohort

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Sensory over-responsivity (SOR), a subtype of sensory processing disorder (SPD), refers to the exaggerated behavioral response to sensory stimuli and affects children of all ages with and without neurodevelopmental disorders.[1,2] SOR is more prevalent and severe among children with autism spectrum disorder (ASD) compared to typically developing children (TD) (effect-size 1.28 (95% CIs 1.11, 1.45) from a meta-analysis of 47 studies) and included in the diagnostic criteria of ASD.[3-5] SOR in the gustatory domain, gustatory sensory over-responsivity (GOR), is expressed as avoidance and/or aversive response to gustatory stimuli.[2] Food selectivity in the ASD population is associated with GOR and brings problematic mealtime behaviors, resulting in family distress in daily life.[6,7] Despite this, there is a paucity of research focused on GOR compared to other SOR domains.

One of the widely used behavioral assessments of sensory processing is the Short Sensory Profile (SSP), a parental report. In addition to a parent-based measure of sensory processing, it is critical to have a standardized assessment for both clinical and research settings. The Sensory Processing 3-Dimensions (SP3D), a structured performance-based measure, was developed to quantify sensory modulation and discrimination in multiple sensory domains in children.[8] This study aimed to compare GOR between the clinician-rated measure, SP3D, and a parental report, SSP, in a pediatric cohort of individuals with and without neurodevelopmental concerns. We hypothesized that GOR scores from the parental report and the clinician-rated measure would correlate in a pediatric cohort. Also, we hypothesized that the GOR group defined by the two measures would agree in this cohort as well. Additionally, we hypothesized that GOR scores and proportions of children with GOR would differ between ASD, SPD, and TD cohorts.

Method

Participants

A total of 93 children (ages 4 to 14 years old) were included using the SPD Consortium Database from the 2 university-affiliated ASD centers and one SPD institution through advertisements in clinics, and local online parent groups. Study inclusion required parental completion of the SSP and the Social Communication Questionnaire (SCQ) as well as the assessment of SP3D by an occupational therapist. To be included in the ASD cohort, children had an SCQ score of ≥15 and exceeded the threshold on the Autism Diagnostic Observation Schedule-2 (ADOS-2). To be included in the SPD cohort, children had one or more of sections or total SSP scores satisfactory for the 'definite difference' in the published norm (cut-off of 2 SDs).[9] Children who did not meet either the ASD or SDP criteria listed above were included in the TD cohort. Furthermore, children had been previously excluded from research studies based on the following criteria: performance IQ less than 70, cases without a reliable informant, and history with congenital and acquired brain injury. Cognitive ability was measured by the Wechsler Intelligence Scale for Children-Fourth edition and the Wechsler Abbreviated Scale of Intelligence. Detailed inclusion and exclusion criteria were published.[10] All parents provided the written consent on behalf of children. The institutional review boards from each institution approved the study (UCSF IRB #13-11320).

Measures

Short Sensory Profile and Gustatory Sensory Over-Responsivity Estimates

The SSP, a short version of the Sensory Profile, is a 5-point Likert scale comprised of 38 items.[9] Based on the performance of 1,037 TD, the norm for typical performance (within 1 SD), probable difference (between 1 SD and 2 SD), and definite difference (beyond 2 SD) score ranges were published. The internal consistency of the taste/smell sensitivity section was 0.90 (Cronbach's alpha) in this study. To extract the SSP GOR score (range of 0-16), we summed 4 items in the taste/smell sensitivity section and inverted the score to make greater scores indicating more severity. GOR+/- group was established using the definite difference scores in the norm.

Sensory Processing 3-Dimensions and Gustatory Sensory Over-Responsivity Estimates

The gustatory domain of the SP3D consists of the Snack time, 6 food item challenges with different textures and tastes, and the Brusha Brusha, brushing challenges to clean the upper and lower sides of teeth. During the subtests, clinicians can choose the observable responses related to typical response, SOR, sensory under-responsivity, and sensory seeking. We chose 3 responses related to SOR (excessive concerns, avoidance or withdrawal behaviors, and adverse responses) among the response options. The presence/absence of each response was transformed into 0 and 1 and summed to make the SP3D GOR score (range of 0-24). The internal consistency of the gustatory domain was 0.72 in this study. We extracted a cut-off SP3D GOR score of 5.96 (>2 SD) from children with typical performance SSP GOR scores. Children with SP3D GOR score above the cut-off score were assigned the GOR+ group.

Analysis Methods and Statistics

Statistical analyses were done using Stata 16.1. Given the skewed distributions by the Shapiro Wilk test, we performed the nonparametric tests in this study. We tested Spearman's correlation between SSP and SP3D GOR scores. Regarding GOR groups, we estimated the inter-test percent agreement between 2 measures (proportion of agreement among the sum of agreement and disagreement) as well as alternative estimates of the Brennan-Prediger and Gwet's AC1. For comparison of GOR scores between cohorts, the Kruskal-Wallis test and Dunn's test as a pairwise comparison were used. We excluded 4 participants with 100% missing SSP GOR items. There were 25 missing observations in the SSP and the SP3D GOR score items from 10 individuals (2.34% of total observations). For these missing values, we performed the chained multiple imputations (n=5) using predictive mean matching with variables of age, sex, and cohorts. Two-sided $p<.05$ was set as a statistical significance.

Results

A total of 89 participants (median age 10 years (interquartile range (IQR) 8-11), male 82% (N=73), median performance IQ 100 (IQR 93-111)) were analyzed to investigate the agreement between the 2 measures and their difference across ASD (n=23), SPD (n=20), and TD (n=40) cohorts. We could not designate 6 children into diagnostic cohorts due to missingness of the SCQ and the ADOS. Both SSP and SP3D GOR scores demonstrated positively skewed distributions (median SSP score 2 (IQR 0-6) vs. median SP3D GOR score 0.5 (IQR 0-2.5)). While the averaged inter-test agreement for GOR group designation was 0.76, suggesting a substantial agreement between the 2 measures, the correlation between the 2 measures was not statistically significant. Additionally, the Brennan-Prediger and Gwet's AC1 were 0.52 and 0.67, indicating moderate agreements. SSP GOR scores differed statistically significantly in the overall comparison ($p<.001$). As a post hoc analysis, we compared the SSP GOR scores and found that the SSP GOR score was significantly lower than the other groups: ASD > TD ($p<.001$) and SPD > TD ($p<.01$). We found a statistically significant difference in the SP3D GOR scores between ASD and TD groups ($p<.05$ in the posthoc comparison).

Discussion

In this study, the correlation of GOR scores was not significant, but 2 measures showed moderate-substantial agreement for defining GOR+/- groups. SSP GOR scores showed high discrimination ability across cohorts, whereas, in the clinician-rated measure (SP3D), only the ASD cohort differed significantly from TD. The GOR+ group had the highest prevalence among the ASD cohort (37-39%), followed by SPD (10-20%) and TD (0-5%) cohorts. Given the limited prior research, this study adds to the literature by performing the comparison of the assessment of GOR by the parental report and the clinician-rated measure and suggesting different purposes of the 2 measures.

The disagreement between the 2 measures could be from differences in settings the behaviors of children are addressed. The parental report is based on behaviors in daily life, whereas the SP3D is performed in clinical settings. Also, 2 measures estimate GOR in different ways. While GOR is quantified by the caregiver's impression in the parental report, in the clinician-rated measure, clinicians observe children's responses to selected items and differentiate the phenotypes based on the responses. In addition, 2 measures have different pros and cons. While the SSP could be easily administered, it addresses GOR broadly and is subject to different standards of caregivers. Cultural, race/ethnic, and socioeconomic factors might play a role on formation of the norm for GOR among caregivers. The clinician-rated measure could be more structured but is dependent on the clinician's specialized experience and skills.

Thus, we might consider the different purposes of the 2 measures since they deliver different information about GOR. While wide distributions of GOR scores in the parental report could help to define children with GOR, the clinician-rated measure could be used to estimate the contributing factors of GOR. For example, ASD populations are known to have more rigidity in thoughts, anxiety in reaction to novel stimuli, as well as poor motor control, and we could find what might be the major factor for GOR through the clinician-rated measure. In addition, contributing factors could differ according to clinical groups of ASD and SPD. Therefore, combinations of the 2 measures would allow us to find children with GOR and apply evidence-based multimodal interventions accordingly. Additionally, we were able to find the distinctive highest GOR estimates in the ASD cohort, which might represent a prominent degree of distress and impact on family members among the ASD population.

There are some limitations to our study. While GOR could differ according to sex, we were not able to balance the number of females and males due to the preponderance of males diagnosed with ASD compared to females. Also, while different socioeconomics, cultural factors, and race/ethnicity might affect the norm for GOR among caregivers, we could not adjust those factors in our study. Additionally, the caregivers who visited the clinics for other reasons would be more likely to volunteer for the study, limiting the transportability of our study result into the TD population in the community. The blinding of presumptive diagnosis was not guaranteed, which could have affected the outcome of the SP3D. Also, because of the limited sample size, we could not separate GOR responses according to the tastes or textures.

Take Home Summary

We compared 2 assessment tools of gustatory over-responsivity (GOR), a parental report and a clinician-rated measure in a pediatric cohort. While 2 measures showed moderate agreement for defining children with GOR, the ASD cohort showed the severest scores in both.

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