Spatial Processing Disorder in Children With Cleft Palate

Le trouble du traitement auditif relié à la spatialité chez les enfants ayant une fissure palatine

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Abstract

Spatial Processing Disorder is manifested as difficulty understanding speech in a noisy environment despite normal standard audiometric results. Rates of Spatial Processing Disorder are significantly higher in children who have a history of otitis media in early childhood, and the prevalence and duration of otitis media in children with cleft palate are significantly higher than the general population. Therefore, this study aimed to determine the prevalence of Spatial Processing Disorder in this vulnerable population. Children with cleft palate aged between 6 and 16 were recruited from a cleft palate clinic. Those with normal audiograms and absence of ear disease, communication disorders, or intellectual disability were included in the study. Eight (40%) of the 20 children who met inclusion criteria were found to have Spatial Processing Disorder using the Listening in Spatialized Noise-Sentences diagnostic standard. Four additional patients were found to have signal-to-noise ratio losses greater than 2 dB from the mean, representing substantial loss in speech intelligibility. Three children underwent remediation using the Listening in Spatialized Noise & Learning program; all saw substantial benefit. Spatial Processing Disorder may be of detriment particularly in school-aged children and is highly prevalent in those with cleft palate. As therapy for this disorder has recently been developed, prompt identification and intervention may improve the learning experience of affected children.

Abrégé

Le trouble du traitement auditif relié à la spatialité se manifeste par une difficulté à comprendre la parole dans un environnement bruyant, et ce, malgré des résultats normaux aux tests audiométriques standards. La proportion d'enfants ayant un trouble du traitement auditif relié à la spatialité est significativement plus élevée chez ceux ayant un historique d'otites moyennes survenues durant les premières années de vie. Ajoutons que la prévalence et la durée des otites moyennes sont significativement plus élevées chez les enfants ayant une fissure palatine que chez les enfants provenant de la population générale. La présente étude visait donc à déterminer la prévalence d'enfants ayant un trouble du traitement auditif relié à la spatialité au sein des enfants ayant une fissure palatine. Des enfants ayant une fissure palatine âgés entre 6 et 16 ans ont été recrutés dans une clinique de fissure palatine. Les enfants ayant des audiogrammes normaux, en l'absence d'un problème otologique, d'un trouble de communication ou d'une déficience intellectuelle, ont été inclus dans l'étude. Huit (40%) des 20 enfants qui respectaient les critères d'inclusion avaient un trouble du traitement auditif relié à la spatialité en utilisant le test *Listening in Spatialized Noise-Sentences*. Quatre autres enfants avaient une perte du ratio signal/bruit supérieure à 2 dB par rapport à la moyenne, ce qui représente une perte importante de l'intelligibilité de la parole. Une intervention utilisant le programme *Listening in Spatialized Noise & Learning* a été offerte à trois des enfants; tous en ont grandement bénéficié. La présence d'un trouble du traitement auditif relié à la spatialité peut porter préjudice aux enfants en étant atteints, et ce, particulièrement à l'âge scolaire. Ajoutons que sa prévalence est élevée chez les enfants ayant une fissure palatine. Étant donné qu'un programme d'intervention a récemment été développé pour ce trouble, une identification et une intervention rapides peuvent améliorer l'expérience d'apprentissage des enfants ayant un trouble du traitement auditif relié à la spatialité.

Spatial Processing Disorder (SPD) can be considered a type of Central Auditory Processing Disorder in which patients with normal standard audiometric test results have listening difficulties (Moore, Rosen, Bamiou, Campbell, & Sirimanna, 2013). Patients with SPD have difficulty attending to streams of speech in noisy environments due to inability to select sounds from a single direction when there is extensive competing background noise (Schneider, 2011). SPD can be debilitating for many patients, but is disproportionately significant in children given their attendance in often-noisy school environments (Mealings, Demuth, Buchholz, & Dillon, 2015).

Recently, a test designed to identify and diagnose SPD has been developed at the National Acoustics Laboratory in Australia (Cameron & Dillon, 2008). This test, the Listening in Spatialized Noise-Sentences (LiSN-S) test, is a virtual, computer-based assessment tool that measures the ability of patients to use spatial cues to differentiate a target talker from competing talkers. Patients with inability to rely on inter-aural cues will predictably perform poorly on spatially separated conditions compared to their counterparts. The effects of non-auditory factors, such as cognitive abilities, attention, and language, are minimized by making the relevant score a "difference" measure between scores on subtests with different acoustic characteristics.

SPD has not been extensively studied, especially in the pediatric population. Preliminary evidence has shown that rates of SPD are significantly higher in children who have a chronic history of otitis media in early childhood (Dillon, Cameron, Glyde, Wilson, & Tomlin, 2012). This suggests that even transient auditory deprivation over time may contribute to the development of SPD due to under stimulation of the central auditory nervous system. In other words, the development of a child's spatial auditory processing abilities could be disrupted by fluctuating hearing levels during critical early developmental periods. Indeed, the prevalence of SPD was found to be significantly higher in Australian Indigenous children, a group with substantially higher rates of otitis media (Cameron, Dillon, Glyde, Kanthan, & Kania, 2014).

Otitis media is common among children with cleft palate. The likelihood of having otitis media by 2 years of age is 97% for children with cleft palate, compared to 60% for the general population (Kuo, Lien, Chu, & Shiao, 2013). Although the incidence of otitis media in this population decreases over time, many children can have chronic Eustachian tube dysfunction and the prevalence of SPD has yet to be examined in children with cleft palate.

The developers of LiSN-S have also created a virtual rehabilitation program designed to improve children's spatial processing abilities and treat SPD. Listening in Spatialized Noise & Learning (LiSN & Learn) employs a series of computer games trained over a period of time, with results showing nearly a full standard deviation of improvement in terms of spatial advantage by the completion of the intervention (Cameron et al., 2014).

This study aimed to (a) assess the prevalence of SPD in children with cleft palate, (b) describe their spatial processing ability, and (c) examine the possibility of SPD rehabilitation using an established remediation program.

Method

This study was approved by the local Research Ethics Board (Izaac Walton Killam Health Centre Protocol # 1020364).

Participants

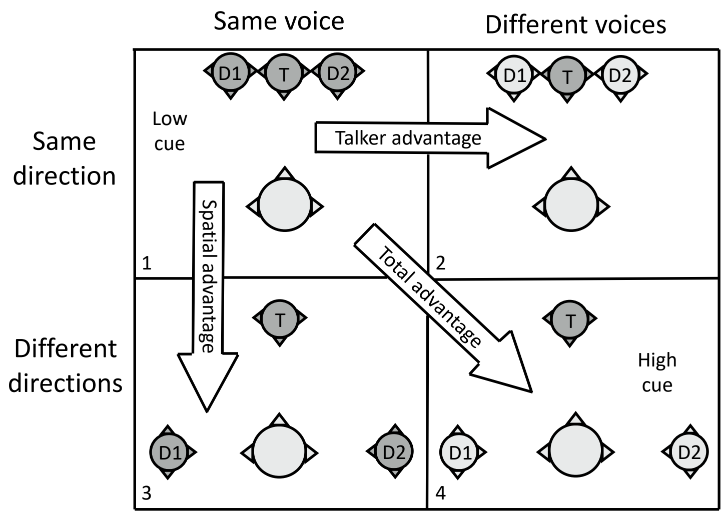
Children with cleft palate (with or without cleft lip) between the ages of 6 and 16 years were recruited from the cleft palate clinic at the Izaac Walton Killam Health Centre in Halifax, Nova Scotia. The age limits were chosen as children under 6 years cannot be reliably tested for SPD using the LiSN-S program (Brown, Cameron, Martin, Watson, & Dillon, 2010). Other inclusion criteria included normal hearing thresholds from 500 Hz to 4000 Hz in both ears and fluency in English. Exclusion criteria included (a) children with intellectual disability, autism spectrum disorder, or with documented, un-treated attention deficit or hyperactivity disorders and (b) abnormal otoscopic examination with the presence of external or middle ear disease.

Medical records of participants were reviewed to obtain demographics, frequency of otitis media diagnoses, and past history of tympanostomy tubes.

Listening in Spatialized Noise-Sentences Test

Participants underwent the LiSN-S test as previously described (Cameron et al., 2009). Briefly, the signal-to-noise ratio (SNR) required to identify 50% of the words in target sentences is established in four conditions that vary in terms of the virtual location of the noise source and the vocal quality of the speaker (**Figure 1**). All sounds are played through a standardized headphone system that stimulates spatialized sound. The target is always presented at 0° azimuth, while competing talkers are presented at 0° (no spatial separation) or at 90° spatial separation. The competing talkers can be the same voice as the target (i.e., no voice cues) or they can be voices different from the target. Using a combination of these parameters, four conditions are created:

Figure 1



The four conditions of the Listening in Spatialized Noise-Sentences test, numbered according to the description in the text in the Method section. Adapted from Cameron, Dillon, Glyde, Kanthan, and Kania (2014). D1 and D2 represent the competing (or distracting) talkers, and T represents the target talker. When D1, D2, and T are the same colour, the voice quality is identical. When D1, D2, and T are different colours, the voice quality is different.

1. Low-cue condition (competition is the same voice at 0° azimuth).
2. Different voices condition (competition is a different voice at 0° azimuth).
3. Spatial separation only condition (competition is the same voice at 90° azimuth).
4. High-cue condition (i.e., competition is a different voice at 90° azimuth).

Subtracting the score for one condition from the score for another results in difference scores. For the purposes of this study, the key difference score is the difference between Condition 3 and Condition 1. This difference score is defined as the *spatial advantage* since the only difference between Condition 1 and Condition 3 is the addition of spatial cues in Condition 3. Less important is the *talker advantage* score, which is defined as the difference between Condition 1 (same voice competition at 0° azimuth) and Condition 2 (different voice competition at 0° azimuth).

Spatial processing disorder was diagnosed using the LiSN-S test if the spatial advantage score was equal to or greater than 2 standard deviations (*SD*) lower than the North American mean (Cameron et al., 2009).

In addition to analyzing data in terms of deviance from the mean, the LiSN-S program also provides the clinician with quantity of SNR loss, if any. For example, if a spatial advantage of 9.0 dB was expected, and the participant's spatial advantage was 6.5 dB, then the participant's SNR loss would be 2.5 dB. Clinically meaningful decrements in SNR advantage were examined, even if the 2 *SD* criterion was not met. A clinically meaningful decrement was defined as an SNR advantage 2 dB below the age-specific mean, since this corresponds to a noticeable difference in a child's ability to listen in a noisy environment (Killion, 2004).

Listening in Spatialized Noise & Learn Training

Children diagnosed with poor performance during the early study period were offered a chance to complete the LiSN & Learn training outside of the hospital environment, either at home or at school. The training protocol has previously been described (i.e., Cameron & Dillon, 2011; Cameron et al., 2014; Cameron, Glyde, & Dillon, 2012). Briefly, children play two auditory training games per day for 5 days each week, for 50 total training sessions or 100 total games (total of 15–20 minutes per day). In all games, the child identifies a word from a target sentence while competing sentences are presented at either 0° azimuth or ± 90° azimuth. Correct answers result in progressively more difficult games in the form of decreasing target voice levels. For incorrect answers, the target voice level is raised.

Data Analysis

Simple descriptive statistics were utilized for quantifying patient characteristics and determining variance from population means for spatial and talker advantages. Two-tailed Student's *t* tests were used for statistical analysis of continuous variables. Correlations were determined using the Pearson correlation coefficient.

Results

Participants

Twenty patients (10 girls) during the study period (October 2015 and October 2016) met inclusion criteria. The median age was 9 years, and all patients had previously repaired cleft palate, with or without cleft lip. The median age at time of cleft palate repair was 11.0 months.

Listening in Spatialized Noise-Sentences Test

Low-cue and high-cue speech reception threshold (SRT) measurements, talker advantage measurements,

Table 1

Raw Listening in Spatialized Noise-Sentences Data for All Patients[1]

Patient	Low-cue SRT	High-cue SRT	Talker advantage	Spatial advantage
01	-0.2	-10.2	2.7	8.6
02	-0.8	-14.6	8.1	13.4
03	1.1	-9.1	2.5	8.3
04	5.1	2.9	2.7	-3.1
05	-2.1	-10.9	3.6	6.1
06	0.6	-14.2	5.6	13.6
07	0.4	-11.1	6.6	8.0
08	1.3	-9.2	4.2	8.1
09	0.0	-6.8	2.9	4.7
10	1.6	-5.2	1.7	4.2
11	2.3	-6.8	3.0	7.8
12	1.9	-8.5	2.5	4.1
13	0.9	-7.5	5.5	6.4
14	7.6	3.6	5.0	4.6
15	0.2	-10.4	1.8	6.8
16	-0.2	-6.8	3.1	5.8
17	0.9	-9.5	4.2	9.6
18	2.4	-7.0	1.1	8.7
19	1.6	-6.2	3.4	6.5
20	-0.1	-8.1	6.1	8.0

Note. [1]All measurements in decibels (dB). SRT = Speech Reception Threshold.

and spatial advantage measurements for all participants are listed in **Table 1**. There were no statistically significant correlations between patient age and low-cue (same voice) SRT, $r(19) = -.50$, $p > .05$; high-cue (different voice) SRT, $r(19) = -.56$, $p > .05$; talker advantage, $r(19) = .45$, $p > .05$; or spatial advantage, $r(19) = .53$, $p > .05$. Similarly, there were no significant gender-related correlations, $r(19) = .33$, .42, -.18, and -.37, respectively, all $p > .05$. There was a strong, significant positive correlation between low-cue SRT and high-cue SRT, $r(19) = .85$, $p < .05$, but not between low-cue SRT and talker advantage or spatial advantage. However, there was a strong, significant negative correlation between high-cue SRT and spatial advantage, $r(19) = -.80$, $p < .05$. The frequency of myringotomy and tympanostomy tube placement did not correlate with any LiSN-S measurements.

Spatial Processing Disorder Diagnosis

Eight patients (40%) were diagnosed with SPD (**Figure 2**). Raw data is available in the Appendix. The median age was 8 years 5 months, with a slight (non-significant) female predominance (62.5%). The characteristics of patients diagnosed with SPD are found in **Table 2**. There was no difference in mean age at time of cleft palate repair between those diagnosed with SPD and those who were not (20.1 vs. 26.0 months, $p > .05$). There was no difference in the mean number of tympanostomy tube procedures required between the two groups (2.2 in those without SPD, 2.9 for those with SPD, $p > .05$).

There were no correlations between variance from the spatial advantage mean and talker advantage scores or talker advantage variance from the mean. The majority of patients were within 2 *SD* of the population mean for their talker advantage measurement (91.7%).

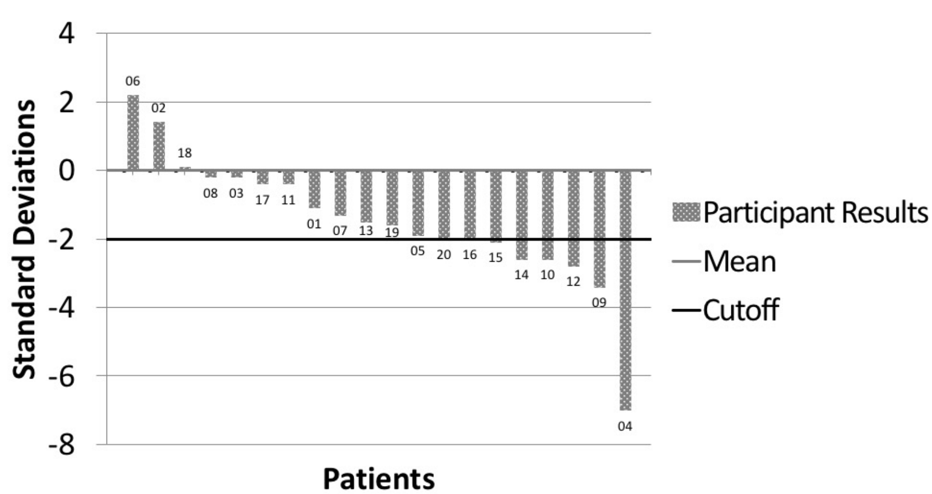
Four patients who were not diagnosed with SPD were found to have a meaningful (> 2 dB) increase in SNR thresholds, resulting in a total of 12 patients (60%) with significant difficulties understanding speech in noise (see **Figure 3**).

Listening in Spatialized Noise & Learn Training

Two children diagnosed with SPD and one child with meaningful increase in SNR thresholds underwent spatial processing remediation with the LiSN & Learn software. Not all eligible participants were offered training due to lack of availability of the remediation software and some did not have the ability to use the program (i.e., lack of appropriate hardware).

The results of remediation training are presented in **Table 3**. All three children had improvement in their

Figure 2



Variance of spatial advantage from the standardized North American population mean for all participants. The cutoff of two standard deviations from the mean was considered significant for the diagnosis of Spatial Processing Disorder. Values above and below the data bars represent the patient ID; for example, the bar to the furthest right is Patient 04. Positive scores represent patients scoring better than the population mean, while negative are worse.

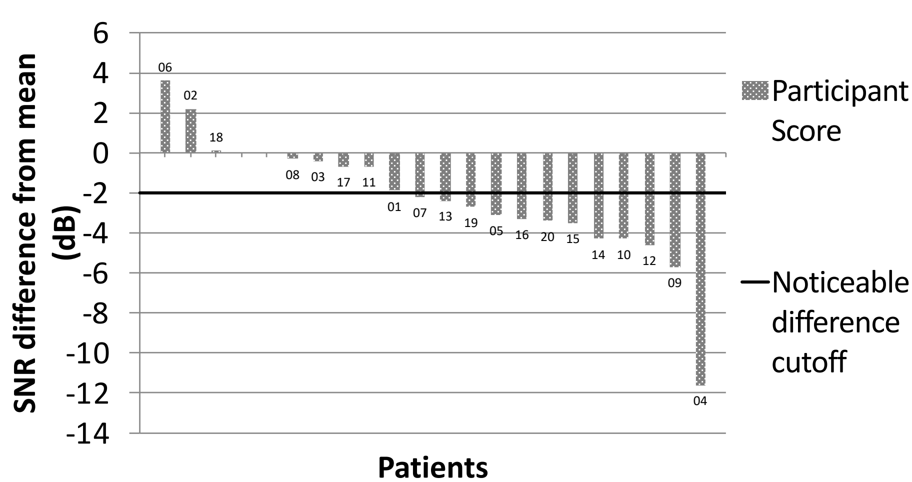
Table 2

Characteristics of Patients Diagnosed With Spatial Processing Disorder

Patient	Age (months)	Diagnosis	Repair	Ear history	Other
04	81	Cleft lip & palate	Yes	M & T x 2	
05	145	Cleft palate	Yes	M & T x 5	
10	82	Bilateral cleft lip & palate	Yes	M & T x 3	
12	91	Cleft lip & palate	Yes	M & T x 3	Loss of alveolar bone graft secondary to trauma
14	97	Unilateral cleft lip & bilateral palate	Yes	OME, COM, No M & T	
15	144	Cleft palate	Yes	M & T x 1	Congenital unilateral blindness
16	104	Submucosal cleft palate	Yes	No M & T	
20	113	Cleft palate	Yes	M & T x 6	

Note. M & T = myringotomy and tympanostomy tubes; OME = otitis media with effusion; COM = chronic otitis media.

Figure 3



Signal-to-Noise ratio differences in the spatial advantage measurement of all participants. The noticeable difference cutoff was defined as two decibels. Values above and below the data bars represent the patient ID; for example, the bar to the furthest right is Patient 04. Positive scores represent patients scoring better than the population mean, while negative are worse.

Table 3

Spatial Advantage Measurements Before and After Remediation With the Listening in Spatialized Noise & Learning Program

Patient	Spatial advantage - pre (dB)	Spatial advantage - post (dB)	Spatial advantage - improvement (dB)	Spatial advantage variance from mean - pre (*SD*)	Spatial advantage variance from mean - post (*SD*)	Spatial advantage variance from mean -improvement (*SD*)
04	-3.10	9.10	12.20	-7.00	0.10	7.10
05	6.10	10.00	3.90	-1.90	0.20	1.70
10	4.20	5.30	1.10	-2.60	-1.70	0.90

Note. dB = Decibels.

spatial advantage and were considered to be within normal range following the intervention.

Discussion

Spatial processing disorder is an under-recognized clinical entity that disproportionately affects children with a history of otitis media. The prevalence of SPD was found to be higher in our study population (40%) than in Indigenous Australian children (7%) who are also thought to be at higher risk for otitis media (Cameron et al., 2014). However, despite having potentially the highest rates of middle ear disease among ethnic groups, rates of otitis media may be higher in the cleft palate population than the Indigenous Australian children. Prevalence of middle ear disease in the Indigenous population has been reported to be as high as 62% (Thorne, 2004), yet rates of otitis media reach nearly 100% in cleft palate patients (Dhillon, 1988). This may explain why the prevalence of SPD was higher in our study. There was a slight female predominance in our study that was not statistically significant. This is likely due to the small population size, as boys are often quoted as having a higher rate of cleft lip and palate than girls.

In addition to the high prevalence of SPD in our patient population, nearly all patients had SNR thresholds that were higher than age-appropriate means. An SNR of only 1 dB above the mean may result in a speech intelligibility loss of up to 17% in noisy environments (Cameron & Dillon, 2007). While SPD may be debilitating for children in learning environments, the loss in speech intelligibility may also be limiting for these children even though they do not formally meet diagnostic criteria for SPD. On the severe end of the spectrum, the patient with 12 dB increase above the mean could be expected to have almost zero sentence recognition in noisy environments.

The lack of correlation between talker advantage and spatial advantage suggests that the scores do not simply reflect higher order language, learning, and communication skills, which would be expected to affect both to a similar degree, but rather that they reflect specific auditory processes. High-cue SRT and spatial advantage measurements had a strong negative correlation, indicating that children with strong spatial listening skills can also take advantage of talker voice differences to improve hearing in noise.

We have also demonstrated the efficacy of LiSN & Learn remediation training in this population, supporting its use and possible future expansion to other patients with SPD. All children showed spatial advantage scores that improved to the normal range following intervention. This is in keeping with the findings of Cameron et al. (2014), in which Indigenous Australian children saw improvements of 1 *SD* on average. One of the children in our study had substantial changes in his or her variance from the population mean, while the other two showed smaller improvements similar to those found in Cameron et al. (2014). The latter participant (Patient 04) was a considerable outlier in terms of both raw spatial advantage score (**Table 1**) and SRT improvement (**Table 3**). Given the small population size in this study, this outlier may have affected correlation calculations. However, many of the LiSN-S correlations are in keeping with expected results and therefore it is unlikely that the outlier caused any substantial statistical aberrations. An earlier randomized, blinded study (i.e., Cameron et al., 2012) reported average SRT reductions of 10 dB. Despite all of the patients in our study having similar *SD* changes as previously reported, only the outlier patient reached the level of dB improvement seen in the trial of Cameron et al.

To our knowledge, the current study is the first to investigate the prevalence of SPD in patients with cleft palate. Despite the small sample size, this study represents all patients from a large geographical area in the Maritime provinces of Canada. Moreover, at the time of testing, all patients had normal otoscopic examinations, normal audiometric testing, and normal tympanometry, so results cannot be attributed to transient differences in middle ear function. Another limitation is that not all participants underwent remediation training due to logistical reasons (i.e., lack of access to the proper equipment to run the LiSN & Learn program), thereby limiting our rehabilitation population size to a small cohort. However, our results are consistent with previous findings that there is improved spatial processing after training.

Future studies should investigate the prevalence of SPD in a larger population of patients with cleft palate, as well as evaluate the effectiveness of the LiSN & Learn program in a larger sample from this population. This study is at low risk for Type II error as the null hypothesis of there being no difference in rates of SPD for patients with cleft palate was rejected. We have limited the Type I error by choosing an alpha of .05 *a priori,* despite the rarity of SPD. Of course, increasing the sample size by a large degree in the future would allow for a stricter *a priori* setting of alpha. It would also be helpful to determine whether training-related improvements in spatial advantage, as found using the LiSN-S, correspond to improvements in functioning in school or in other real world noisy environments.

In summary, nearly half of cleft palate patients in our study population, despite medical and surgical intervention, were found to have spatial processing difficulties and therefore could have significant hearing issues when in a noisy environment. Given the improvements demonstrated by the use of the LiSN & Learn training program, albeit in a limited sample, these difficulties may be amenable to remediation. This is the first study to investigate the prevalence of SPD in patients with cleft palate and the first to demonstrate the potential remediation of SPD in this group.

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Appendix

Raw Variance Data From Listening in Spatialized Noise-Sentences Testing

Patient ID	Low-cue SRT		High-cue SRT		Talker advantage		Spatial advantage		
	Variance from mean LCSRT score	Variance from mean in *SD*	Variance from mean HCSRT score	Variance from mean in *SD*	Variance from mean TA score	Variance from mean in *SD*	Variance from mean SA score	Variance from cutoff score	Variance from mean in *SD*
01	0.9	-0.9	2.2	-1.1	-4.2	-1.9	-1.9	1.4	-1.1
02	0.7	-0.8	-1.3	0.6	0.4	0.1	2.2	5.6	1.4
03	1.0	-1.0	0.6	-0.3	-2.0	-0.9	-0.4	2.9	-0.2
04	4.8	-5.0	12.3	-5.9	-1.5	-0.7	-11.6	-8.2	-7.0
05	-1.9	1.9	-0.3	0.2	-1.6	-0.7	-3.1	0.2	-1.9
06	1.3	-1.4	-2.5	1.2	-0.6	-0.3	3.6	6.9	2.2
07	1.2	-1.3	0.8	-0.4	0.2	0.1	-2.2	1.2	-1.3
08	1.0	-1.0	0.1	-0.1	0.1	0	-0.3	3.0	-0.2
09	1.0	-1.0	5.4	-2.6	-3.8	-1.7	-5.7	-2.3	-3.4
10	1.3	-1.3	4.2	-2.0	-2.5	-1.1	-4.3	-0.9	-2.6
11	2.0	-2.1	2.6	-1.3	-1.2	-0.6	-0.7	2.7	-0.4
12	1.8	-1.9	1.3	-0.6	-1.9	-1.0	-4.6	-1.3	-2.8
13	0.9	-0.9	2.5	-1.2	1.2	0.4	-2.4	0.9	-1.5
14	7.6	-7.9	13.7	-6.6	0.2	0.1	-4.3	-1.0	-2.6
15	1.2	-1.2	1.7	-0.8	-4.8	-2.2	-3.5	-0.2	-2.1
16	0	0	3.6	-1.7	-2.0	-0.9	-3.3	0	-2.0
17	1.8	-1.9	2.5	-1.2	-2.4	-1.1	-0.7	2.7	-0.4
18	2.2	-2.3	2.6	-1.3	-3.3	-1.5	0.1	3.4	0.1
19	1.8	-1.9	4.3	-2.1	-1.8	-0.8	-2.7	0.6	-1.6
20	0.3	-0.3	2.8	-1.3	0.6	0.3	-3.4	0	-2.0

Note. SRT = Speech Reception Threshold; LCSRT = Low Cue Speech Reception Threshold; HCSRT = High Cue Speech Reception Threshold; TA = Talker Advantage; SA = Spatial Advantage.