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Examining the Speech Intelligibility of Individuals With Oromandibular Dystonia Receiving Botulinum Toxin: A Series of Cases



Examiner l'intelligibilité de la parole des individus ayant une dystonie oromandibulaire et recevant des injections de toxine botulique : une série de cas

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Abstract

Oromandibular dystonia is a focal dystonia affecting the facial, lingual, and labial musculature. Oromandibular dystonia can result in a hyperkinetic dysarthria with associated reductions in speech intelligibility. Botulinum toxin A injections are the gold standard in the therapeutic management of oromandibular dystonia. Unfortunately, there is a sparse empirical literature that has examined changes to speech intelligibility in individuals receiving botulinum toxin A therapy. In this preliminary study, we measured the speech intelligibility of 10 individuals with oromandibular dystonia at two time points over the course of a therapeutic botulinum toxin A injection cycle. Intelligibility was assessed using the Sentence Intelligibility Test and a conversational speech task. Four listeners rated sentence intelligibility and conversational intelligibility via visual analogue scaling. Changes to speech intelligibility over the course of the botulinum toxin A treatment cycle were analyzed using a series of cases. The speech intelligibility of one individual with lingual oromandibular dystonia demonstrated significant increases to speech intelligibility over the course of the treatment cycle. The remaining nine participants demonstrated relative stability in speech intelligibility scores over the course of the treatment cycle. It appears that for jaw opening, jaw closing, and mixed presentations of oromandibular dystonia, botulinum toxin A injections did not significantly reduce or improve speech intelligibility. Our preliminary results provide a rationale for examining speech intelligibility and the response to botulinum toxin A based on the type and location of oromandibular dystonia in larger scale study.

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Abrégé

La dystonie oromandibulaire est une dystonie focale affectant la musculature faciale, linguale et labiale. La dystonie oromandibulaire peut entraîner une dysarthrie hyperkinétique, celle-ci associée à une réduction de l'intelligibilité de la parole. Les injections de toxine botulique de type A sont considérées comme étant la prise en charge de référence pour la dystonie oromandibulaire. Malheureusement, il existe peu d'études empiriques qui ont examiné les changements dans l'intelligibilité de la parole des individus recevant des injections de toxine botulique de type A. Dans la présente étude préliminaire, nous avons mesuré l'intelligibilité de la parole de 10 individus ayant une dystonie oromandibulaire, et ce, à deux moments au cours d'un cycle d'injection de toxine botulique de type A. L'intelligibilité a été évaluée à l'aide du *Sentence Intelligibility Test* et d'une tâche conversationnelle. Quatre auditeurs ont évalué l'intelligibilité de la parole dans les échantillons de phrases et de conversation, et ce, grâce à une échelle visuelle analogique. Un devis de série de cas a été utilisé pour analyser les changements observés au niveau de l'intelligibilité de la parole. Une augmentation significative de l'intelligibilité de la parole au cours du cycle de traitement a été observée pour un individu ayant une dystonie oromandibulaire affectant la musculature linguale. L'intelligibilité de la parole des neuf autres participants est demeurée relativement stable au cours du cycle de traitement. Il semble que les injections de toxine botulique de type A n'augmentent pas ou ne diminuent pas l'intelligibilité de la parole des individus ayant une dystonie oromandibulaire affectant les muscles responsables de l'ouverture ou de la fermeture de la mâchoire, ou encore, affectant plusieurs muscles du bas du visage (p. ex. linguaux, labiaux et/ou faciaux). Les résultats de cette étude préliminaire supportent la réalisation d'études de plus grande envergure pour examiner l'effet de la toxine botulique de type A sur l'intelligibilité de la parole, et ce, en fonction du type et de la localisation de la dystonie oromandibulaire.

Oromandibular dystonia (OMD) is a focal dystonia affecting the mouth and facial regions (Tan, 2004). It consists primarily of forceful involuntary contractions of the facial and lingual musculature. These contractions may either be sustained or repetitive. Other terms for OMD are orofacial-buccal dystonia, jaw dystonia, lingual dystonia, cranial dystonia, and adult-onset facial dystonia (Schneider & Hoffman, 2011). In some cases, OMD occurs with blepharospasm or involuntary contractions of the eyelids. This condition is called Meige's syndrome (Lee, 2007). Although the exact cause of dystonia is unknown, it has been recognized as a disease involving basal ganglia (Kaji, 2003; Shanker & Bressman, 2012; Tsui, 2005). Additionally, the DYT1 gene has been noted to play a role in the onset of dystonia (Tagliati, Pourfar, & Bressman, 2005; Tsui, 2005).

The basal ganglia refer to a group of nuclei in the central nervous system that plan and execute motor movements (Mink, 2003). Lesions isolated to the putamen and globus pallidus of the basal ganglia are the most frequently associated with dystonia (Bhatia & Marsden, 1994). The extent of basal ganglia involvement in dystonia remains poorly understood; however, it has been hypothesized that dystonia results from reduced firing of neurons within the globus pallidus interna. This decreased activity of the globus pallidus interna neurons leads to incomplete inhibition of competing motor movement patterns. Reduced inhibition of these surrounding motor patterns can lead to the involuntary contraction of neighbouring muscles (Mink, 2003).

The DYT1 gene has been implicated in causing the greatest number of primary dystonias that have been genetically researched (Tagliati et al., 2005). A deletion of a GAG sequence in the DYT1 gene leads to dystonia (Tagliati et al., 2005). The DYT1 gene encodes torsinA, a protein that is involved in vesicle fusion and cytoskeletal dynamics (Tagliati et al., 2005).

Because OMD involves abnormal contraction of the facial muscles, it may produce difficulty in mastication and swallowing (Bhidayasiri, Cardoso, & Truong, 2006; Lee, 2007). It may also lead to difficulties in opening and closing the mandible and controlling the lingual and labial musculature. These difficulties can result in dysarthria, defined as

a collective name for a group of speech disorders resulting from disturbances in muscular control over the speech mechanism due to damage of the central or peripheral nervous system. It designates problems in oral communication due to paralysis, weakness, or incoordination of the speech musculature (Darley, Aronson, & Brown, 1969a, p. 246).

The type of dysarthria most frequently associated with OMD is a slow hyperkinetic dysarthria in which the most affected aspects of speech production are articulatory in nature and include imprecise consonants, distorted vowels, and irregular articulatory breakdowns (Duffy, 2013).

OMD is among the most challenging types of dystonia to treat (Jankovic, 2004). Because of the various clinical presentations and severities of OMD, it has become a challenge among clinicians to properly diagnose this condition (Balasubramaniam, Rasmussen, Carlson, Van Sickels, & Okeson, 2008). There are many available clinical treatments for OMD. The most common and well-tolerated treatment for OMD is chemodenervation. This is achieved via localized injection of botulinum toxin into the affected muscles. In rare cases, neurosurgical surgical intervention, such as deep brain stimulation of the globus pallidus internus is also possible, but a less common approach (Capelle, Weigel, & Krauss, 2003). The most common oral medications for OMD are anticholinergic drugs such as trihexylphenidyl, dopaminergics, dopamine receptor blockers, carbamezapines, and baclofen (Tsui, 2005). Oral baclofen has been shown to be commonly used in OMD (Jankovic, 2005; Tan, 2004; Tsui, 2005), and has been reported to be effective in 20% of patients with OMD (Tsui, 2005). In general, treatment of OMD using pharmaceuticals has been reported to be unremarkable and reports of side effects have been high (Cultrara, Chitkara, & Blitzer, 2004; Jankovic, 2004; Tsui, 2005).

Botulinum toxin A (BoNT-A), known commercially as Botox® (Allergan, Inc. Irvine, CA, USA) and Xeomin® (Merz Pharmaceuticals, Germany), has been used in the treatment of OMD (Batla, Stamelou, & Bhatia, 2012; Bhattacharyya & Tarsy, 2001; Cultrara et al., 2004; Teemul, Patel, Kanatas, & Carter, 2016). Not only are BoNT-A injections effective in alleviating symptoms of dystonia, but research has also shown that use of BoNT-A can be effective in improving the quality of life with patients with OMD, including the subdomains of social support and physical health (Bhattacharyya & Tarsy, 2001). Similarly, it has also been found that BoNT-A injections can improve domains of activity and participation, as well as improving social, emotional, and vocational aspects of general well-being (Dykstra, Adams, & Jog, 2007).

In contrast, the effect of BoNT-A injections on speech intelligibility in OMD has a sparse empirical literature, and therefore, is poorly understood in comparison to more studied outcome measures such as quality of life (see Bhattacharyya & Tarsy, 2001; Teemul et al., 2016). This is unfortunate because dysarthria can be a disabling

consequence of OMD (Tan & Jankovic, 1999). Dykstra, Domingo, Adams, and Jog (2015) was one of the first groups of researchers to conduct a study that examined ratings of speech intelligibility and self-rated communicative effectiveness in individuals with OMD over the course of a BoNT-A injection cycle. The results of their study found no significant overall group differences in sentence intelligibility or self-rated communicative effectiveness over the course of a treatment cycle of 3 months (Dykstra et al., 2015). This 3-month re-injection schedule is the standard protocol for re-injection in OMD because BoNT-A has a wearing off cycle of approximately three months (Blitzer & Sulica, 2001). To our knowledge, there has been no report of permanent stabilization of symptoms or cumulative effects of BoNT-A over prolonged periods of treatment (Bakheit, Liprot, Newton, & Pickett, 2012; Colosimo, Tiple, & Berardelli, 2012).

Although Dykstra et al. (2015) did not find significant group differences in speech intelligibility over the course of the BoNT-A treatment cycle in their participants with OMD, it is possible that individual differences may have been present based on the type and/or location of OMD that was not revealed through the analysis of aggregated intelligibility data.

Current Study

This preliminary study examined, on an individual, case-by-case basis, the speech intelligibility of 10 participants with various presentations of OMD over the course of a single BoNT-A treatment cycle. Using case reports, this study aimed to evaluate if therapeutic BoNT-A injections produced differential changes to speech intelligibility based on the type and/or location of OMD. Although this study is preliminary and exploratory in nature, it is hypothesized that those individuals with primarily lingual involvement will derive a greater benefit to speech intelligibility following BoNT-A therapy as compared to individuals with other presentations of OMD such as jaw-opening or jaw-closing. This hypothesis is driven by a case study that reported improved speech intelligibility following BoNT-A injections of an individual with lingual dystonia (Dykstra et al., 2007). Furthermore, previous literature suggests that tongue control is more strongly related to speech intelligibility than jaw or lip control in individuals with neuromotor disorders (Weismer, Yunusova, & Bunton, 2012). Finally, in previous work examining the efficacy of BoNT-A injections on OMD, it has been found that jaw-opening OMD was associated with less functional improvement and higher complication rates after BoNT-A injections than jaw-closing OMD (Tan & Jankovic, 1999; Teive et al., 2012). Taken together, these studies suggest possible differential effects of BoNT-A on speech intelligibility based on the articulator primarily affected by OMD.

The data from the current study and from Dykstra et al. (2015) come from a larger clinical study in which the same set of participants was asked to complete several intelligibility tests and questionnaires. The results of these tests are reported across the two articles, depending on the specific questions being addressed. Further, the current study uses a subset of the listeners recruited in Dykstra et al. (2015). The advantage of using the same raters is that the results of both studies are comparable. One participant was excluded in the current paper because that participant provided many outlier responses for the measures we reported. As such, it was difficult to ascertain whether these anomalous results were because of true perceptual differences or if that participant did not understand the visual analogue scaling (VAS) task or make an effort to give accurate responses.

In Dykstra et al. (2015), researchers sought to explore potential relationships between sentence intelligibility judged by transcription with patient-reported self-ratings of communicative effectiveness. Because the transcription-based intelligibility scores were quite high in the first published study, researchers sought to determine if VAS estimates of intelligibility would reveal more about the speech intelligibility of people with OMD over the course of their treatment cycle. The current study aims to address that question by examining VAS estimates of sentence intelligibility and conversational intelligibility, neither of which were explored in the 2015 paper.

Evaluating sentence intelligibility using VAS and comparing it with VAS-rated conversational intelligibility are novel and important contributions to the literature because daily communication is almost entirely composed of conversational, spontaneous speech. Furthermore, because we had a heterogeneous sample of OMD types, it was important to explore the potential differences in intelligibility outcomes on a case-by-case basis.

Method

Participants

Participants with OMD. Ten participants with OMD participated in this study. **Table 1** provides a summary of participant demographics. Participants were diagnosed with OMD by a neurologist specializing in movement disorders (M. J.). The same neurologist also administered therapeutic BoNT-A (Botox® or Xeomin®) injections to participants as part of their routine clinical care. Participants were tested just prior to their pre-scheduled therapeutic BoNT-A injections occurring on an ongoing 3-month schedule, when effects are believed to have worn off (Blitzer & Sulica, 2001).

Table 1

Demographic Information of Participants With Oromandibular Dystonia

Participant case	Sex	Age	OMD duration (years)	Years receiving BoNT-A	Frequency of injection (months)	Type of OMD	Injection site and type of BoNT-A
1	M	69	4	3	3	Meige's (labial)	Orbicularis oris: 10u total h/s (Xeomin®)
2	F	78	2	3 months	3	Jaw opening	R&L lateral pterygoid: 30u total, R&L digastric: 40u, f/s (Botox®)
3	F	60	10	8	3	Lingual	Genioglossus: 15u total, R&L digastric: 40u total, f/s (Botox®)
4	F	69	21	21	3	Lingual, labial, jaw closure	R&L pterygoid: 30u total, R&L digastric: 10u total, f/s (Xeomin®)
5	M	78	13	11	3	Labial, jaw closure	Orbicularis oris: 60u total, R&L masseter 40 units total, f/s (Botox®)
6	M	56	4	4	3	Jaw opening, jaw closure, lingual	R&L lateral pterygoid: 140u total, R&L digastric: 40u total, tongue: 30u total, f/s (Botox®)
7	M	80	23	22	3	Meige's (jaw opening, jaw closure)	R&L lateral pterygoid: 120u total, R&L digastric: 30u total, f/s (Xeomin®)
8	M	68	8	3	3	Jaw closure	R&L masseter: 30u total, medial pterygoid: 30u total, f/s (Botox®)
9	F	67	5	4	3	Meige's (labial)	R&L digastric: 10u total, R&L pterygoid: 20u total, f/s, Orbicularis oris: 5u, h/s (Botox®)
10	M	44	3	1	3	Meige's (labial, jaw closure)	R&L masseter: 40u total, medial pterygoid, 40u total, f/s (Botox®)

Note. BoNT-A = Botulinum toxin A; OMD = oromandibular dystonia; R = right; L = left; u = units; f/s = full strength; h/s = half strength.

A speech-language pathologist (A. P.) with over 15 years of experience with dysarthria determined that participants demonstrated hyperkinetic dysarthria associated with OMD. Participants were recruited if they were receiving BoNT-A injections to manage their symptoms of OMD, presented with dysarthria, and had no other speech impairments other than those resulting from OMD. Participants also reported that they were not receiving speech therapy and

were not taking any other medications that could impact motor function. In total, there were 6 men and 4 women (age range = 44–80 years, *M* = 66.9) recruited to participate, with an average OMD onset of 13.8 years.

This study was approved by the Health Sciences Research Ethics Board at Western University (Research Ethics Board approval #101658) and occurred over two

testing sessions. The experimenter obtained informed consent before both testing sessions.

Listeners. Four naive individuals were recruited to participate in this study as listeners. These listeners were young adults, 20–23 years of age ($M = 21$ years). All listeners were native English speakers; had no known speech, hearing, or neurological impairments; and had no familiarity with dysarthric speech. All listeners passed a 30 dB HL hearing screening bilaterally at 500, 1000, 2000, and 4000 Hz before participating.

Procedure

The speech intelligibility of participants with OMD was assessed over two experimental sessions. The first experimental session, referred to as the *pre-BoNT-A* condition, occurred immediately before participants received their routinely scheduled BoNT-A injections. This also corresponded to approximately three months after participants' last BoNT-A injections, except for one participant who was de-novo. The second experimental session, referred to as the *post-BoNT-A* condition, occurred approximately four to six weeks after participants received their BoNT-A injections to correspond to peak therapeutic effectiveness (Blitzer & Sulica, 2001).

Speech intelligibility. All recordings were done in a quiet testing room. Each participant wore a headset microphone (AKG C520) that was placed 6 cm from his/her mouth and connected to a digital audio recorder (Zoom H4n) that recorded the participant's speech at a 16 bit and 44 kHz sampling rate. Each recording session lasted approximately 10 minutes.

Participants completed sentence and conversational intelligibility tasks during each experimental condition (pre- and post-BoNT-A). Sentence intelligibility data was obtained using stimuli from the Sentence Intelligibility Test (SIT; Yorkston, Beukelman, & Hakel, 1996), which has been found to be a valid and reliable tool for assessing speech intelligibility in dysarthric speakers (Yorkston, Strand, & Kennedy, 1996). The SIT is comprised of lists of 11 unique and randomly generated sentences ranging from 5–15 words in length. Each participant read aloud a different and randomly generated list of SIT sentences and he or she was audio recorded for later analysis of speech intelligibility.

Conversational speech samples were obtained by asking each participant to talk about a familiar topic while being audio recorded. Open-ended questions were used to elicit spontaneous responses. Example questions included "What do you do for a living?" "What are your hobbies?" and "Tell me about your last vacation." Participants were asked different

questions in the pre-BoNT-A and post-BoNT-A conditions to ensure that their responses were unrehearsed.

After each session, SIT recordings from each participant with OMD were combined into a single excerpt using Praat (Boersma & Weenink, 2013). Each participant with OMD therefore had two SIT excerpts, corresponding to the two experimental conditions. Conversational intelligibility samples were created from a continuous segment of spontaneously generated speech lasting about 30–45 seconds in duration. Selection of conversational samples were not blinded to treatment condition, but in order to maintain consistent quality for all samples, the following criteria were applied: (a) segments had to have minimal to no filler words present, (b) segments needed to be 30–45 seconds in duration (i.e., splicing together several shorter segments to create a longer segment was not permitted), and (c) segments had to have no extraneous sounds (e.g., adjusting the chair or microphone, coughing, yawning) or further probing by the experimenter (e.g., "Can you tell me more about that?"). All SIT excerpts and conversational speech samples were then numbered and compiled into playlists generated by Windows Media Player (version 12). All playlists were counterbalanced and randomized so that each listener was presented with SIT and conversational recordings from each OMD participant in a different order. Each participant with OMD produced 11 SIT sentences in the pre-BoNT-A condition and another 11 SIT sentences in the post-BoNT-A condition, creating a total of 22 SIT sentences. Additionally, each participant produced 30–45 seconds of conversational speech pre- and post-BoNT-A, creating 60–90 seconds of conversational speech across both experimental conditions. Across both treatment conditions and both tasks, participants produced a total of approximately 3–3.5 minutes of speech.

After all speech samples were compiled and edited, each listener completed a single listening session lasting approximately 90 minutes in a quiet laboratory wherein free-field presentation of speech samples were played at a comfortable listening level via M-Audio speakers (AV 40) placed approximately 0.6 metres (24 inches) away. During this listening session, listeners used VAS to rate the speech intelligibility of the recorded sentences and conversational speech samples obtained from participants with OMD during pre- and post-BoNT-A experimental conditions.

Speech intelligibility was rated by listeners on a 100mm visual analogue scale with the anchors labeled *0% intelligible* on the left and *100% intelligible* on the right side of the VAS. Listeners were presented with SIT sentences and conversational speech samples and

they were required to indicate the level of intelligibility by drawing a hatch mark along the 100mm line corresponding to how intelligible they perceived the speech sample to be. Speech intelligibility was measured as the distance in millimetres from the left end of the scale to where the hatch was drawn and was expressed as a percentage (i.e., 83 mm = 83% perceived intelligibility).

To determine whether participants with OMD experienced significant changes in intelligibility, we followed the guidelines of the Assessment of Intelligibility of Dysarthric Speech (Yorkston & Beukelman, 1984), from which the SIT was derived, that stated that sentence intelligibility must change by a minimum of 8.6% to be considered a clinically significant difference.

Results

Reliability

Inter-rater and intra-rater estimates of reliability were calculated for both sentence and conversational intelligibility tasks. Scores from each listener for each intelligibility task were measured against each other to obtain inter-rater reliability values. All four listeners re-measured 10% of data to determine intra-rater reliability.

Table 2 summarizes the intra-class correlation and Cronbach’s alpha values in obtaining inter-rater and intra-rater reliability values.

The intra-class correlation value obtained for overall inter-rater reliability was .910 with 95% confidence intervals between .854 and .948. This correlation coefficient demonstrates an excellent reliability measure among listeners for our sentence and conversational speech intelligibility measures. Furthermore, we found moderate intra-rater reliability within measurements of each listener. The intra-class correlation was .847 with 95% confidence intervals between .373 and .965.

Case Reports

A summary of sentence and conversational intelligibility scores and measured intelligibility change pre- and post-BoNT-A injections for each participant is presented in **Table 3**.

Applying the threshold of 8.6% change in intelligibility as a benchmark of clinical significance to both our sentence and conversational ratings, a significant increase to both sentence and conversational intelligibility over the course of a single BoNT-A treatment cycle was observed in only one (Case 3) of our nine participants. This was our only participant that presented with an isolated lingual dystonia. The other participants we studied presented with either jaw opening (Case 2), jaw closing (Case 8), labial (Cases 1 and 9), or mixed (Cases 4, 5, 6, 7, and 10) presentations of OMD. **Figure 1** shows percentage change between treatment conditions whereby participants are classified as having lingual dystonia only (Panel A), mixed case involving lingual dystonia (Panel B), or the absence of involvement (Panel C).

For most of our participants studied, relative stability was observed in speech intelligibility across the treatment cycle with the exception of Case 10 who demonstrated a relatively large increase in conversational intelligibility (+16.25%) from pre- to post-testing despite minimal changes to sentence intelligibility ratings (+2.88%).

Of the five participants (Cases 2, 4, 6, 8, and 9) who demonstrated decreased conversational intelligibility, four (Cases 2, 4, 6, and 8) presented with dystonic symptoms involving the jaw. While this is an interesting pattern of results, it is not conclusive evidence and we do not suggest that BoNT-A injections worsen conversational intelligibility because Cases 5 and 9 also presented with jaw involvement but did not demonstrate the same pattern. This result warrants careful consideration in a future study.

Across the 10 participants, both sentence and conversational estimates of intelligibility remained relatively stable over the course of a single BoNT-A injection cycle.

Table 2		
Summary of Inter-Rater and Intra-Rater Estimates of Reliability for Sentence and Conversational Intelligibility Tasks		
	Intra-rater reliability	Inter-rater reliability
Intraclass Coefficient Correlation	.847	.910
Cronbach’s Alpha	.915	.910

Table 3

Mean Sentence and Conversational Intelligibility Scores of Participants with Oromandibular Dystonia and Their Corresponding Intelligibility Change Pre- and Post-BoNT-A.

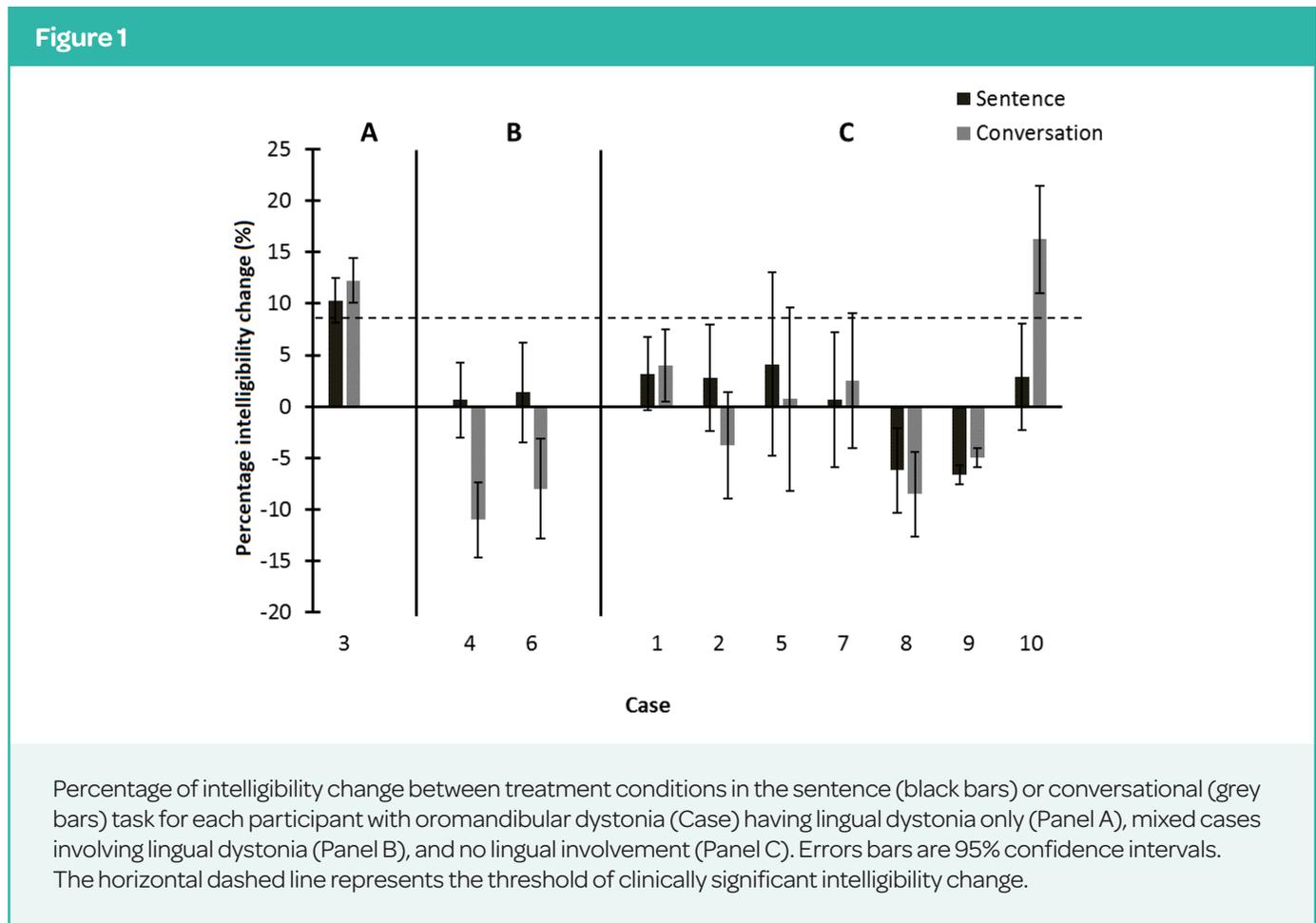
Case	Type of OMD	SIT – VAS		Direction and magnitude of change (%)	Conv		Direction and magnitude of change (%)
		M Pre (SD)	M Post (SD)		M Pre (SD)	M Post (SD)	
1	Meige's (labial)	89.31 (10.35)	92.50 (6.26)	+3.19	84.75 (9.57)	88.75 (11.93)	+4.00
2	Jaw opening	91.25 (6.97)	94.06 (5.17)	+2.81	93.25 (5.74)	89.50 (9.26)	-3.75
3	Lingual	82.81 (16.88)	93.13 (8.29)	+10.32	82.00 (9.42)	94.25 (5.19)	+12.25
4	Lingual, labial, jaw closure	77.75 (10.25)	80.63 (13.62)	+2.88	33.00 (19.51)	49.25 (21.09)	+16.25
5	Labial, jaw closure	76.25 (13.84)	76.88 (9.05)	+0.63	82.00 (7.39)	71.00 (13.49)	-11.00
6	Jaw opening, jaw closure, lingual	75.06 (5.20)	79.19 (9.49)	+4.13	79.13 (6.49)	79.88 (18.12)	+0.75
7	Meige's (jaw opening, jaw closure)	46.94 (21.25)	48.31 (20.77)	+1.37	55.63 (17.61)	47.63 (23.02)	-8.00
8	Jaw closure	78.63 (12.05)	79.31 (13.24)	+0.68	71.50 (17.46)	74.00 (15.06)	+2.50
9	Meige's (labial)	95.44 (3.70)	89.25 (11.44)	-6.19	88.50 (8.58)	80.00 (12.75)	-8.50
10	Meige's (labial, jaw closure)	87.88 (6.85)	81.25 (10.37)	-6.63	84.25 (8.66)	79.25 (7.46)	-5.00

Note. BoNT-A = Botulinum toxin A; OMD = oromandibular dystonia; SIT-VAS = Sentence Intelligibility Test measured using Visual Analog Scaling; Conv = Conversational intelligibility.

Mean VAS sentence intelligibility ratings increased post-BoNT-A by 1.31% (median = 2.09%, range = 16.94%) and conversational estimates of intelligibility decreased by 0.05% (median = -1.5%, range = 27.25%). Eight participants (Cases 1, 2, 3, 4, 5, 6, 7, and 10) demonstrated increased sentence intelligibility over the course of the injection cycle and five of these eight participants (Cases 1, 3, 5, 7, and 10) also demonstrated increased conversational intelligibility over the course of the injection cycle. Cases 2, 4, and 6 demonstrated decreased conversational intelligibility despite improved sentence intelligibility. The remaining

two participants (Cases 8 and 9) who demonstrated decreased sentence intelligibility also demonstrated decreased conversational intelligibility over the course of a single injection cycle. None of the participants were rated to have increasing conversational intelligibility but decreasing sentence intelligibility scores.

A Pearson correlation analysis was conducted to determine if there was a relationship between change in intelligibility scores and the amount of BoNT-A injected, as well as change in intelligibility scores to each other. Only



participants receiving full-strength BoNT-A were included in this analysis, therefore data from Case 1 was excluded. The correlation between SIT change and units of BoNT-A was not significant ($r = .11, p = .78$), neither was the correlation between conversational intelligibility change and units of BoNT-A ($r = -.08, p = .84$). There was also no significant correlation between the percentage of intelligibility change in SIT and conversational tasks ($r = .62, p = .08$).

Discussion

Oromandibular dystonia is a focal dystonia primarily affecting the muscles of the lips, jaw, and tongue. The motor speech disorder associated with OMD, a slow hyperkinetic dysarthria, is primarily characterized by imprecise consonant articulation, vowel distortion, and irregular articulatory breakdown (Darley, Aronson, & Brown, 1969b). Dysarthria is known to have global impacts on speech production affecting parameters such as intelligibility, prosody, voice quality, and speech rate (Kent, 2000).

In the current study, we presented a case series of 10 individuals with varying presentations of OMD and

associated hyperkinetic dysarthria and we examined both sentence and conversational speech intelligibility at two time points over the course of a single BoNT-A injection treatment cycle. Nine out of 10 participants were experienced with BoNT-A injections since they had been receiving BoNT-A for several years prior to participating in the study, while one participant was de-novo. It remains unclear whether long-term use of BoNT-A results in stabilization of dystonic symptoms over time (Colosimo et al., 2012). Bakheit et al. (2012) investigated the possibility of developing neutralizing BoNT-A antibodies that block the action of BoNT-A. In this study, patients had a minimum of 10 consecutive treatment cycles spaced at least 3 months apart, and none of them developed antibodies. Importantly, over the course of the study BoNT-A treatment was found to be at least partially beneficial in 97% of cases, suggesting that the therapeutic effects of BoNT-A are present after prolonged periods of treatment.

Our rationale for examining intelligibility using a case series approach was not only to further our knowledge of intelligibility deficits in this under-studied clinical population,

but it also served to provide preliminary data to justify, in a larger scale study, the examination of the differential effects of BoNT-A on speech intelligibility based on type and/or location of OMD.

Task-specific changes in intelligibility were found in Case 10 in which there were large increases in conversational intelligibility but relative stability in sentence intelligibility. This result is somewhat puzzling, but the large standard deviation associated with this participant suggests a high degree of variability in how our listeners rated conversational intelligibility in this participant. Inspection of his sentence intelligibility scores versus conversational intelligibility scores also show a significant overall discrepancy in speech intelligibility ratings when reading sentences from the SIT (pre = 77.75%, post = 80.63%) versus his intelligibility in a conversational task (pre = 33.00%, post = 49.25%), regardless of BoNT-A injections. Anecdotally, this task effect difference was very prevalent during testing. One potential explanation is that reading tasks (i.e., sentence intelligibility) provide an external model or cue which serves to decrease demands placed on the basal ganglia with regard to the planning and the execution of motor speech movements and the additional demands involved in planning spontaneous speech movements (Kempler & Van Lancker, 2002). Kempler and Van Lancker (2002) demonstrated this task effect by showing that lower intelligibility scores are associated with spontaneous speech versus read speech in a participant with dysarthria associated with Parkinson's disease.

It is also possible that there is a task-specific dystonic response when reading that serves to disrupt dystonic movements during reading versus in spontaneously produced conversation. This mechanism can be considered similar to a *geste antagoniste*. A *geste antagoniste*, also referred to as a sensory trick, is a voluntary maneuver such as chewing or laughing, by which participants can temporarily decrease dystonic symptoms (Blitzer & Sulica, 2001). It has been suggested that the relief of dystonic symptoms occurs through the activation of different sensory pathways (Giladi, 1997). Perhaps relief of dystonic symptoms may also be achieved by activating different pathways involved in planning motor movements, such as those involved in reading as opposed to generating spontaneous speech. Case 10 is particularly intriguing and his task specific changes in intelligibility require more detailed examination in a future study.

We gathered anecdotal evidence that many of our participants felt a great deal of disablement due to their speech production difficulties associated with OMD that

were not fully alleviated by their therapeutic BoNT-A injections. Despite these impressions our participants expressed, and in conjunction with our data, we do not consider our results to be an indicator that BoNT-A injections are not effective in our participant group. The pre-BoNT-A intelligibility scores of most of our participants were already quite high. Yorkston and Beukelman (1984) defined mild dysarthria as having intelligibility scores in the range of 95%. In our sample, four of our participants with OMD had intelligibility scores above 90%; therefore, it is possible that we are observing ceiling effects in intelligibility.

It has been suggested that dysarthrias caused by chronic conditions, as is the case in the current study, cannot be resolved solely by medical interventions alone (Kent, 2000). Therefore, relying on BoNT-A treatment exclusively to manage speech-related deficits caused by OMD may not be a realistic expectation. A recent systematic review that examined the use of BoNT-A as a treatment for OMD discussed the highly variable outcomes experienced by individuals with OMD pre- and post-BoNT-A treatment and emphasized the need for further research on use of BoNT-A treatment in OMD (Comella, 2018). The inconsistent effects of BoNT-A injections on speech intelligibility in the current paper support the conclusions of Comella (2018) and reinforce the importance of evaluating the suitability of BoNT-A in OMD.

The treatment of OMD with BoNT-A injections has focused on the improvement of dystonic muscle contractions, pain management (Cultrara et al., 2004; Esper, Freeman, & Factor, 2010; Teive et al., 2012), orofacial esthetics, chewing and mastication, and health-related quality of life (Bhattacharyya & Tarsy, 2001; Teemul et al., 2016). Speech-related outcomes appear to have a secondary priority to those factors listed above.

Although there is no cure for OMD, behavioural therapy in conjunction with BoNT-A therapy may be helpful in the management of dystonic symptoms that impair speech intelligibility (Yorkston et al., 1996). Common behavioural interventions for dysarthria include, but are not limited to, articulation exercises, breath control exercises, and rate control techniques such as the use of a pacing board (Yorkston et al., 1996). There is also a demand for a combination of both behavioural and medical interventions; however, the efficacy of combining interventions has not been empirically examined and is thus still poorly understood (Kent, 2000). Furthermore, by combining behavioural interventions with BoNT-A treatment, the management of dystonia and its related symptoms can be customized to individuals based on the subtype of OMD with which

they present as well as the severity of symptoms.

Clinical Implications

Based on our preliminary results discussed above, it appears that speech intelligibility may show differential patterns of response to BoNT-A based on the location of the dystonia and the articulator(s) affected. More specifically, our results provide preliminary support to our hypothesis that those with lingual presentations of OMD may derive more benefit to intelligibility from BoNT-A injections than other presentations such as jaw-opening, jaw-closing, labial, or mixed OMD. These interpretations are evidenced by Case 3, the only participant in our sample to present with lingual dystonia only and who demonstrated improvements in both sentence and conversational intelligibility above the threshold of clinical significance. Our results may be explained by Weismer et al. (2012) who asserted that the tongue is the most influential articulator for intelligibility as compared to other articulators such as the jaw. It may be that muscles of the tongue are more responsive to BoNT-A injections than muscles with greater mass such as the masseter (i.e., jaw closure dystonia). Additionally, it appears that there are significant individual differences in changes to intelligibility evident in even our small sample of participants as shown by the range of improvement and decline in intelligibility scores across task (see range of y-axis values in **Figure 1**), which suggests an individualized approach to management.

Taken together, our results provide useful clinical implications for speech-language pathologists. First, we must be mindful of the potential differences to speech intelligibility in response to BoNT-A injections rather than viewing this medical intervention as a one-size-fits-all approach. Moreover, we must pair medical interventions (i.e., BoNT-A injections) with other strategies for supporting effective communication and tailor intervention to our clients' needs and potentially based on the type and location of OMD. By providing additional communication support, such as the provision of strategies that can be used to improve effectiveness of communication, we can ultimately ensure that the clients we serve have opportunities for meaningful communicative interactions and for participation in a variety of contexts and roles that involve communication.

Lastly, we must be cognizant of the impact of BoNT-A therapy on other aspects of an individual's functioning such as pain management, improvement to swallowing and mastication, facial esthetics, and overall impact on quality of life outcomes. Only by taking a

multi-faceted approach will we gain a comprehensive understanding of the benefits of BoNT-A injections, despite observing relatively stable speech intelligibility over a single treatment cycle for many of the individuals with OMD we assessed.

Limitations

While this study provides preliminary data to support the hypothesis that subtypes of OMD may show differential benefits to speech intelligibility over the course of a BoNT-A treatment cycle, the findings of this study should be interpreted with caution due to some study limitations. The first limitation relates to our small sample size, which limits the generalizability of our results.

Second, nine of our 10 participants were already receiving ongoing BoNT-A injections before participating in the current study. Although it has been demonstrated that effects of BoNT-A wear off after approximately three months (Blitzer & Sulica, 2001), we were not able to determine with certainty if our participants had experienced the complete wearing off effects and had returned to their baseline intelligibility. Perhaps our participants would have demonstrated a greater change to their speech intelligibility scores over the course of a treatment cycle and perhaps even demonstrated greater improvements to their intelligibility scores if we were able to obtain a true return to baseline. This would have been possible if BoNT-A injections were delayed for a longer period. Future studies may wish to extend the injection cycle to ensure BoNT-A has completely worn off when obtaining baseline speech intelligibility measures.

Third, injections were not performed under electromyography guidance so there is a possibility that the precise location of dystonic activity may not have been injected. Future studies may wish to use electromyography-guided injections. Lastly, although the SIT is usually rated via orthographic transcription, we opted for visual analogue scaling to provide a more consistent measure between sentence and conversational speech samples. Conversational or spontaneously generated speech samples have higher face validity compared to sentence intelligibility measures because the majority of everyday communication occurs spontaneously; therefore, speech elicited in conversational tasks is the most naturalistic (Kent, Weismer, Kent, & Rosenbek, 1989). Future studies may want to include orthographic transcription and acoustic measures such as the second-formant slopes to provide a more detailed description of intelligibility in this clinical population. Second-formant slopes have been shown

to correlate well with measures of intelligibility involving single-word identification (Kent, Kent et al., 1989) and may be considered as a measure of motor involvement in speech (Kim, Weismer, Kent, & Duffy, 2009).

Future Directions

Due to our small sample size, this study has not yielded definitive conclusions of how BoNT-A may produce differential changes to speech intelligibility based on OMD subtypes. However, we believe the trends in our preliminary data present a clear direction for future work. A larger scale, multi-centre study is warranted to systematically assess how therapeutic BoNT-A injections impact speech production across various subtypes of OMD. In addition to measuring speech intelligibility from a perceptual approach, it would also be valuable to assess changes in acoustic and kinematic parameters in a pre- and post-injection paradigm.

Further, since BoNT-A injections are the “gold standard” of treatment for OMD (Tan & Jankovic, 1999), it would be of interest to evaluate and understand how BoNT-A impacts other aspects of functioning to get a comprehensive understanding of the benefits of BoNT-A. Ideally, this would include a detailed evaluation and assessment of the effects of BoNT-A injections on mastication and swallowing, speech intelligibility, and facial esthetics, as well as self-ratings of health-related quality of life, communication-related quality of life, and communicative participation. This information could help to provide much needed objective and patient-reported outcome data to determine the overall benefit of BoNT-A injections.

Further investigation of the relationship between severity of symptoms, location of dystonia, speech rate, and total number of words produced in the conversational task is also valuable to understand and characterize how location of dystonia affects speech production. Gathering this information in a larger scale study but also on an individual basis during clinical interactions could inform treatment planning and tailor the appropriate management of OMD. This can be achieved by examining the relative importance of each aspect of impaired functioning on an individual's life and if BoNT-A injections provide the desired benefit.

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Elicited and Spontaneous Determiner Phrase Production in French-Speaking Children With Developmental Language Disorder



Production induite et spontanée de syntagmes nominaux (*determiner phrase*) chez les enfants francophones ayant un trouble développemental du langage

KEYWORDS

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Abstract

We contrast elicitation and spontaneous speech data in French-speaking children with developmental language disorder and controls, with a focus on determiner phrase and gender agreement. Eight French-speaking children with developmental language disorder and age-matched or language-matched controls were compared on an elicitation task for complex noun-phrases with one or two adjectives (e.g., *la petite maison verte* 'the small green house') and a spontaneous speech sample of 200 utterances containing determiner phrases. Elicitation and spontaneous speech data revealed different profiles in French children with developmental language disorder compared to controls: elicitation tasks revealed specific difficulties with adjective agreement as well as high levels of global error, while spontaneous speech revealed mostly omission and substitution errors, often on determiners. Ultimately, both approaches to evaluating language abilities are complementary, but elicitation tasks might be the most useful tool for rapid identification of difficulties with determiner phrases and agreement in young French-speaking children.

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Nous comparons les données recueillies dans des tâches de production induite avec celles provenant d'un échantillon de langage spontané chez des enfants francophones ayant un trouble développemental du langage et des enfants de groupes contrôles, en mettant l'accent sur les syntagmes nominaux (*determiner phrase*) et leur accord en genre. Les productions de 8 enfants francophones ayant d'un trouble développemental du langage, provenant d'un échantillon de 200 énoncés spontanés contenant des syntagmes nominaux ou recueillies dans des tâches de production induite de syntagmes nominaux complexes contenant un ou deux adjectifs (p. ex. *la petite maison verte*), ont été comparées avec celles de participants contrôles appariés sur l'âge ou sur les habiletés langagières. Les données recueillies dans les tâches de production induite et celles provenant de l'échantillon de langage spontané ont révélé que le profil des enfants francophones ayant un trouble développemental du langage différait de celui des enfants des groupes contrôles. Les résultats aux tâches de production induite ont révélé que les enfants ayant un trouble développemental du langage avaient des difficultés spécifiques avec l'accord des adjectifs et produisaient, de façon générale, un plus grand nombre d'erreurs. Les résultats provenant de l'échantillon de langage spontané ont révélé que les enfants ayant un trouble développemental du langage faisaient principalement des erreurs d'omission et de substitution, et ce, souvent avec les déterminants. Ultimement, les deux approches utilisées sont complémentaires pour évaluer les habiletés langagières des enfants. Néanmoins, les tâches de production induite semblent davantage utiles pour identifier rapidement les difficultés qu'ont certains enfants francophones avec les syntagmes nominaux et leur accord.

Developmental language disorder (DLD; Bishop, Snowling, Thompson, Greenhalgh, & CATALISE-2 consortium, 2017; in Québec, *trouble développemental du langage*) is found in approximately 7% of the population in Québec, Canada (Ordre des orthophonistes et audiologistes du Québec, 2014). DLD describes children who exhibit persistent language difficulties in the absence of obvious neurological impairment, social deprivation, or low cognitive skills (Leonard, 2014; Ordre des orthophonistes et audiologistes du Québec, 2004). This disorder can impair the lexicon, phonology, morphology, syntax, morphosyntax—for example, accusative clitics (Grüter, 2005), gender agreement (Roulet-Amiot & Jakubowicz, 2006), and verb inflection (Royle, St-Denis, Mazzocca, & Marquis, 2017)—and pragmatics in the expressive or receptive spheres (Leonard, 2014). However, French-speaking children with DLD are somewhat difficult to identify because in the preschool years they show almost no omission or substitution errors in spontaneous speech (Elin T. Thordardottir & Namazi, 2007), but can have reduced syntactic structure repertoires as compared to age-matched controls (e.g., in French noun-phrases; Royle & Stine, 2013). The relative lack of French indicators for this disorder has led to a search for specific markers for DLD in French. Our focus is on grammatical gender agreement and noun phrase structures, or determiner phrases (DPs) because they are acquired early and reveal language production difficulties in children with DLD.

French Agreement and Acquisition of the Determiner Phrase

French exhibits subject-verb, number, and person agreement as well as gender agreement, which is especially prevalent in the DP. Masculine is the default gender but French contains equal numbers of masculine and feminine nouns. Many elements in French, such as determiners, relative pronouns, pronoun clitics, adjectives, and other structures agree with the noun with which they are co-referential, either in gender, in number, or both. However, agreement processes are not always morphologically transparent. For example, pronouns and determiners have plural forms that are underspecified for gender (e.g., *les* def.pl 'the', the accusative clitic *les* 'them', or *leur* dat.cl.pl 'to them') and many adjectives and past participles have invariable forms: compare variable *brun/e* [br̥œ̃/bʁyn] 'brown.m/f' to invariable *jaune* [ʒœ̃n] 'yellow,' two of our stimulus items.

Longitudinal and cross-sectional corpora showed that typically developing French children produce their first variable feminine adjectives between ages 1;8 and 2;4, and all children show strong mastery of determiner

agreement with mean scores of 96% (Valois & Royle, 2009; Valois, Royle, Sutton, & Bourdua-Roy, 2009). Few variable adjectives, however, were found in these corpora—only eight different lemmas were found in the first, and five in the second. Children can produce agreement in elicitation tasks between ages 4 and 7, depending on the structure. Determiner agreement is reliably elicited before age 5, while adjective agreement becomes stable around age 6 or later (Roulet-Amiot & Jakubowicz, 2006). This last phenomenon might be linked to item-based lexical learning (Royle & Valois, 2010) as some young typically developing children (aged 3;0 and 3;4) are unable to comprehend specific adjectives' feminine forms when asked to provide, e.g., a green frog, *Donne moi la grenouille verte* [vɛʁʁ] 'Give me the.f frog green.f'; response, *Il n'y en a pas* 'There are none', even though they understand the masculine form *vert* [vɛʁʁ] (Royle & Valois, 2010). Brain imaging data show that neurotypical French adults and children aged 7 or 8 years process agreement in similar ways (Fromont, Royle, & Steinhauer, 2015).

Gender Agreement and Determiner Phrases in Children With Developmental Language Disorder

Some researchers (i.e., Rice & Oetting, 1993; van der Lely, 1998) have proposed that DP agreement is a relative strength in English-speaking children with DLD. However, children with DLD have been found to show agreement difficulties in many languages including Dutch, English, Icelandic, Italian, Swedish (see Leonard, 2016, for a short review), and French (Gopnik, 1990; Parisse & Maillart, 2007; Pizzioli & Schelstraete, 2008). Further, determiner substitutions and omissions have been found in Spanish (Bedore & Leonard, 2001) and Swedish (Hansson, Nettelblatt, & Leonard, 2003). Difficulties with genitive (e.g., *hundens mat* 'the dog's food'), plural inflection, indefinite articles (e.g., *en hund* 'a dog'), and determiner + adjective + noun structures (e.g., *dom smutsiga flickorna* 'the dirty girls') are also observed in Swedish (Leonard, Salameh, & Hansson, 2001). In English, children with DLD have difficulties with DP syntax, including comprehending specificity marked by definite and indefinite determiners or genitive structures such as *that/those bear's/bears' balloon/s* (Ramos, 2000). Russian-speaking children with DLD up to age 10 have been shown to exhibit persistent gender agreement errors on adjectives (Tribushinina & Dubinkina, 2012), while Rakhlin, Kornilov, and Grigorenko (2014) observed difficulties in children with DLD up to age 15 in judging feminine Russian gender, as opposed to masculine gender, which is the default.

However, a coherent picture of determiner adjective and preposition use in the noun phrase has not emerged

from the literature on French. For example, in a 20-minute spontaneous-speech sample, Le Normand, Leonard, and McGregor (1993) observed no differences in determiner omissions between a mean length of utterance (MLU) matched group (aged 3–3;6) and French-speaking children with language impairment (aged 4–6 years) on a small number of utterances ($M = 29$). On the contrary, Parisse and Maillart (2007) observed a higher percentage of syntactic structure errors with determiners and prepositions in four French-speaking children with DLD aged 3–7 years as compared to MLU-matched children, based on short spontaneous-speech samples collected over two to four 20-minute encounters. However, no error description is presented.

Gopnik (1990) observed errors on determiners and adjectives in a corpus of 70 utterances for a 9-year-old French-English bilingual child with DLD. When describing pictures, he alternated *le* and *les* for plural targets (**le/les marshmallows* ‘the.sg/pl marshmallows’), omitted determiners (*Après tire fleur* ‘After [he] throws [a/the] flower’), and made gender errors on determiners and adjectives in spontaneous speech (*Il prend *un *gros respiration* ‘He takes a.m big.m breath’, breath is feminine in French). In contrast, using 45-minute spontaneous speech samples—the number of utterances is not specified—Paradis and Crago (2001) observed no difficulties in spontaneous production of determiners and prepositions among 10 French-speaking children with DLD (average age: 7;6), their age-matched controls, and their MLU-matched controls (average age: 3;3). Pizzioli and Schelstraete (2008) computed determiner omissions in a sentence-production task focusing on verb argument structure with participants aged 8;1 to 13;0, controls matched for age, and controls matched on sentence production abilities (aged 5;6–6;4). They found that omissions were higher in the DLD group—5–17%, depending on conditions, exact numbers are not provided—than language-matched (2.5%) and age-matched control groups (no errors). Roulet-Amiot and Jakubowicz (2006) elicited adjective production in French children with DLD aged 6;10 to 12;6 and control groups aged 4 and 6 years. In their sentence completion task (e.g., Here the elephant is wearing ___ [a green shirt]), children with DLD showed higher error rates ($M = 27\%$), mostly on feminine adjectives, than both control groups (4-year-olds: $M = 19\%$; 6-year-olds: $M = 5\%$).

These authors also evaluated “comprehension” errors using a semantic categorization task (e.g., Can you eat that?) with auditory presentation of grammatically correct and incorrect DPs (e.g., *un *nouvelle balai* ‘a.m new.f broom.m). Despite globally higher categorization error rates in participants with DLD (i.e., 9.5% versus 5.8%

in 6-year-olds and 4.1% in adults), no ungrammaticality or slowing effects were found in children with DLD faced with ungrammatical structures. The authors concluded that these children are sensitive to gender agreement during language comprehension and that it is only the extra processing load involved in producing the structures that causes errors. However, it is unclear whether the task was appropriate for this question; at least for these types of structures, the comprehension task could easily be carried out without agreement checking. Furthermore, the absence of reaction time differences between grammatical and ungrammatical structures could point to an *insensitivity* to gender agreement errors in children with DLD. Finally, as far as we can tell, the authors did not control stimuli for any psycholinguistic factors such as age of acquisition or word-internal morphological structure.

What can we conclude from these mixed results? It appears that French-speaking children can exhibit at least some level of error on agreement in determiners and adjective production and do not always resolve these issues, as shown by studies including children aged 6–13 years. The difficulties appear to be more marked on elicitation tasks, but are occasionally also observed in longer spontaneous speech corpora, and this might depend on their age. We believe that elicited agreement is a fruitful domain for study in French child language, as spontaneous production in young children does not systematically distinguish preschool French-speaking children with DLD from their peers on morphosyntactic abilities. For example, Elin T. Thordardottir and Namazi, (2007) found virtually no differences on a number of grammatical morphemes they studied. Spontaneous speech corpora might therefore over- or under-evaluate their language abilities (Eisenbeiss, 2011; Royle & Elin Thordardottir, 2008; Steel, Rose, Eadie, & Thornton, 2013). Further, because of low adjective variety in production even when children appear to master them (Royle & Valois, 2010), elicitation might be a more efficient approach to study them. We thus used the DP elicitation paradigm developed by Royle and Valois, as well as spontaneous corpus analyses, to establish the usefulness of either approach for the identification of agreement difficulties in French and other difficulties that might arise in DP structures (e.g., syntax, determiner production, and omission of obligatory elements).

Current Study

This study compared two approaches to the investigation of noun-phrase structures in French. The first, an elicitation task using puzzles, was used to evaluate the production and comprehension of adjective and determiner agreement, as well as syntactic structures—

mainly Adj-Noun or Noun-Adj order—in simple (e.g., *La maison brune* ‘The.f. house brown.f.’) and slightly more complex noun-phrases (e.g., *La grande maison brune* ‘The.f big.f. brown.f house’). The second approach was used to evaluate noun-phrase production in the spontaneous speech corpus. This approach allowed for comparison of similar structures across both tasks, but also other types of DP structures not elicited with the puzzle props, that emerged in the corpus.

The major research question was whether children with DLD have difficulties producing DPs with appropriate agreement and other syntactic and morphosyntactic structures that are common in them, especially word-order and prepositional structures. A secondary question was to compare elicitation and spontaneous speech tasks for the identification of difficulties in DP structures in children with DLD.

Because younger French-speaking children with DLD have not systematically shown obvious difficulties producing adjective agreement or other morphosyntactic structures in spontaneous speech, but have shown more difficulties in controlled elicitation contexts, we expected elicitation tasks to highlight agreement processing difficulties on adjectives and determiners, and difficulties with DP structure, in children with DLD. We expected spontaneous speech corpora to reveal difficulties less saliently, more specifically on adjectives because adjectives are rare and not varied in spontaneous speech corpora for typically developing French-speaking children (see Valois & Royle, 2009, and Valois et al., 2009, for transversal and longitudinal corpus analyses), and because determiner errors (omissions and commissions) were not consistently found in the studies reviewed above. However, with more variety in structures produced (e.g., determiners, prepositions, adjectives, complex nouns phrases with expansions), the corpus has the potential to provide us with interesting data that go beyond the constrained set of items used in controlled experiments. We thus expected the corpus to reveal other difficulties children with DLD might have with DP structures.

Method

Participants

Participants were residents of the province of Québec, Canada, had French-speaking parents, and were exposed to French at least 80% of the time at home or in daycare. They had no history of autism, neurological disorder, hearing loss, learning disabilities, or other medical conditions that could impair language development. Participant characteristics are presented in **Table 1**. Parents signed a consent form for their child’s participation. All children gave oral consent to participate.

Children with Developmental Language Disorder.

Nine monolingual French-speaking children from Montréal and the Sherbrooke region (three girls and six boys, two of which were twins: D5 and D6) were recruited through specialized services for children with language disorders (i.e., a research hospital and a summer camp). They had been labelled as having *dysphasie* (the term for DLD at the time) by a certified speech-language pathologist, between 9 months and up to 3 years prior to testing. They had maintained their diagnosis as determined by response to intervention. Based on parental questionnaires, all children were monolingual and raised in monolingual environments. One child was excluded because he was significantly older than the controls. The remaining eight were between 5 and 6 years of age ($M = 5;7$ years, $SD = 0;4$) and had a mean length of utterance in words (MLUw) of 4.4 ($SD = 0.97$).

Control groups. In total, 25 control participants were included in this study. Two of our four puzzles (puzzles 3 and 4, see below) had A and B versions. Control children only saw one of the boards for these two puzzles (only A or B) in order to mitigate potential fatigue. We realized that task fatigue was not an issue and all but two children with DLD were presented with the complete set of stimuli (D2 and D4 saw only the A versions). Thus, control matching for puzzle analyses necessitated double pairings. Each child with DLD was matched with two typically developing peers, having done either A or B versions of tasks 3 and 4, on biological sex, parental education, non-verbal memory IQ, and either (a) age: the AGE group, matched for age within two months always to the advantage of children with DLD, or (b) MLUw: the MLU group based on a sample of 100 utterances (a subset of the 200 utterances used for the corpus analysis). All control children were from a larger pool ($N = 151$) of children recruited from the greater Montréal area. Because two controls were needed for every pairing on the puzzle tasks, but only one for the spontaneous-speech sample, half of the children matched for the task were randomly selected for the spontaneous speech sample comparisons (details are provided in **Table 1**). As matching child-by-child made it somewhat difficult to find perfectly matching pairs on all measures, even among our large sample of controls, we reused data from some control children (e.g., N114) as they matched with more than one child with DLD. In total, 12 AGE controls (8 in spontaneous speech, marked with \diamond in **Table 1**), and 13 MLU controls (8 in spontaneous speech) were chosen. No significant differences were found between DLD and control groups on parental education or MLUw scores (see Elin Thordardottir et al., 2005, for similar findings).

Table 1

Participant Characteristics

	Age (months)	MLUw	MLUm	Sex	MEM IQ	EVIP (centile score)	EDUC (years)
DLD group^a							
1. D2	65	4.02	5.18	F	109	71	22
2. D3	70	3.75	4.91	M	96	87	14
3. D4	60	2.88	3.79	M	90	<1	mv
4. D5	67	4.39	5.79	M	96	75	11
5. D6	67	5.38	7.17	M	90	30	11
6. D7	69	4.17	5.50	M	106	11	12
7. D8	69	4.64	5.10	F	74	14	11
8. D9	71	6.02	8.06	F	53 ⁺	15	14
Mean ^c	67.25	4.41	5.69		89.25	37.9	13.57
SD	3.49	0.97	1.35		18.16	34.23	3.95
AGE group^{a,b}							
1. N55 ⁺	63	4.70	6.10	F	106	79	15
2. N50 ⁺	69	4.25	5.37	M	122	97	14
2. N109	69	5.13	6.93	M	90	81	14
3. N42 ⁺	58	4.59	6.47	M	103	50	14.5
4. N20 ⁺	67	4.49	5.92	M	100	99	13.5
4. N114	66	4.92	6.83	M	93	98	14
5. N20 ⁺	67	4.49	5.92	M	100	99	13.5
5. N114	66	4.92	6.83	M	93	98	14
6. N101 ⁺	69	5.27	7.31	M	103	97	14.5
6. N151	72	7.38	9.38	M	103	81	12
7. N127 ⁺	68	3.65	5.57	F	112	64	12
7. N69	67	5.65	7.31	F	125	99	12
8. N9 ⁺	71	4.11	5.77	F	100	66	10.5
8. N8	71	5.28	6.96	F	106	96	13.5
AGE Puzzle Controls (n = 14)							
Mean ^c	66.64	4.92	6.62 [*]		104	86 ^{**}	13.36
SD	5.17	0.88	1.02		10.10	16.20	1.26
AGE Corpus Controls (n = 8)							
Mean ^c	66.5	4.44	6.05		105.75 [*]	81.38 [*]	13.44
SD	4.14	0.47	0.61		7.72	19.41	1.5

MLU group ^{a,b}							
1. N63 [‡]	49	3.67	4.74	F	74	80	20
2. N96 [‡]	45	3.87	4.64	M	106	68	10
2. N36	43	3.75	4.61	M	90	77	13.5
3. N103 [‡]	38	2.68	3.53	M	96	52	14.5
4. N41 [‡]	56	4.28	5.71	M	109	50	13
4. N141	46	4.34	5.66	M	106	79	14.5
5. N115 [‡]	49	5.61	7.51	M	125	50	15
5. N129	68	5.14	6.79	M	131	86	14
6. N80 [‡]	46	4.21	5.66	M	122	73	12
6. N96	45	4.11	5.82	M	106	68	10
7. N92 [‡]	43	4.21	5.56	F	143	62	15.5
7. N71	58	4.15	5.19	F	96	66	10
8. N3 [‡]	44	6.15	7.77	F	87	30	14
8. N11	51	6.21	8.12	F	106	95	16
MLU Puzzle Controls (n = 14)							
Mean ^c	48.64 ^{***}	4.46	5.81		106.93	66.86 ^{**}	13.71
SD	7.65	0.99	1.32		18.50	17.08	2.72
MLU Corpus Controls (n = 8)							
Mean ^c	46.25 ^{**}	4.34	5.64		107.75 [*]	58.13	14.25
SD	5.28	1.09	1.43		22.22	15.91	2.93

Note. AGE = controls matched on age; DLD = children with developmental language disorder; MLU = controls matched on mean length of utterance. mv = missing value; EDUC = mean parental education; EVIP = *Échelle en images Peabody* [French Peabody Picture Vocabulary Test receptive vocabulary]; F = female; M = male; MEM IQ = Leiter memory subtest (Associated Pairs and Forward Memory subtests, [‡] this child was non-compliant on the MEM IQ; however, she appeared to have normal cognitive abilities in all respects); MLUw = mean length of utterances in words; MLUm = mean length of utterances in morphemes.

^aNumbers refer to children with DLD and their matched controls. ^bParticipants marked with a [‡] were included in the spontaneous speech analysis. ^cT tests between DLD and control groups

* $p < .05$, ** $p < .01$, *** $p < .001$.

AGE-matched controls did not differ with the DLD group on age, but the MLU-matched group did. Significant differences were observed between DLD and both AGE-matched and MLU-matched control groups on receptive vocabulary scores, except for the MLU-matched sub-group in the corpus analysis who nonetheless showed a trend for higher scores than the DLD group ($p = .076$). Memory IQ scores were not significantly different in groups matched for the puzzle comparisons, but were significantly higher in both MLU- and AGE-matched groups for the corpus comparison.^{1,2}

Materials

We used four puzzles containing images varying in size or colour to elicit targeted adjectives and nouns (see Royle & Valois, 2010, for details). Participants had to name the pieces they wanted to manipulate. The first puzzle tested colour adjective vocabulary: six colour dots with no gender information, half variable and half invariable, usually produced in the default masculine, although this was not scored for target production. The second depicted eight

¹ This might be due to the fact that the (presumably) non-verbal Leiter screen loads heavily on linguistic abilities: it uses cards with visual cues such as a crib, a plate, a boot, etc., that children must reorganize in specific sequences. See also Miller and Gilbert (2008) for a comparison of two non-verbal intelligence tasks and DLD.

² Note that one child with DLD (D9) was non-compliant during the IQ task and was an outlier that significantly reduced group scores. Excluding this child and her controls for the comparison show that there are significant differences in memory IQ between AGE-matched and DLD groups ($p = .037$), and that the comparison between MLU-matched and DLD groups is no longer significant but shows a trend ($p = .056$).

items: four nouns (half masculine) varying in size, which forced appropriate use of gender-marked determiners and adjectives *small* and *big*, both variable. The third contained four nouns that varied in colour, with 24 items over two versions, A and B, half with variable adjectives (e.g., *the brown duck* vs. *the white duck*) forcing the use of gender-marked determiners and adjectives. The fourth puzzle tested DPs with both colour and size characteristics and contained 48 items over two versions, A and B, using all adjectives and nouns from previous puzzles, all forcing the use of gender-marked determiners and adjectives. Each A and B version of the second and third puzzles contained half of the stimulus items to minimize task fatigue. Appendix A includes the full set of items for version A and Appendix B presents examples of visual stimuli. Images were printed on the backgrounds of puzzle boards and on insert wood pieces. All adjectives and nouns used were early-acquired and high-frequency in oral French based on oral language frequencies in New, Pallier, Ferrand, and Matos (2001) and age-of-acquisition norms from Trudeau, Frank, and Poulin-Dubois (1999; see Royle & Valois, 2010, for details).

Procedure

All participants took part in two 1.5-hour experimental sessions within 2 weeks, during which they completed a hearing screening, the *Échelle de vocabulaire en images Peabody*, a receptive vocabulary assessment (Dunn, Thériault-Whalen, & Dunn, 1993), the Leiter Memory Screen (Associated Pairs and Forward Memory subtests; Roid & Miller, 1996), the puzzles, and an audio-video recorded spontaneous-speech sample of the child playing with a standard set of toys (house, circus-train, plasticine, bus). The language samples were used to evaluate structures of interest and to establish MLU scores. Non-directive speech was used to interact with the children while they played. A hearing threshold in both ears above 20 at 500 Hz, or 15 at 1000, 2000, and 4000 Hz resulted in the child being excluded from the experiment. All tasks were run by trained researchers most often in a sound-proof recording lab equipped with an observation post for parents. This took place at the *Centre de recherche Marie-Enfant*, Montréal, Québec. Some children with DLD were tested at their day camp in Sherbrooke, Québec, over 2 days in a quiet room.

During the puzzle barrier tasks, the experimenter installed a screen between herself and the child to avoid pointing. Puzzle boards were presented sequentially in ascending order of difficulty. Two practice stimuli were presented to the child before each puzzle, with target models provided orally (e.g., "If you wanted this piece, you could say 'give me the pink horse.' I am behind the screen and cannot see the puzzle so you must tell me with words which piece you want"). The

child was then encouraged to ask the experimenter for the piece he or she wanted to put on the puzzle. If he or she successfully performed the first puzzle, the experimenter moved on to the second one, and so on. During the second meeting, a comprehension task was run using the same materials and procedure while reversing experimenter and child roles. All procedures were approved by the *Centre de recherche CHU Ste-Justine* Institutional Review Board (#2167) and the *Université de Montréal Comité d'éthique à la recherche en santé* (#14-034-CERES-P(2)) committees.

Analyses

Puzzles. Target responses and error types were noted. Responses were scored in three ways. The first scored the full appropriate (target) response, which in the first puzzle was simply naming the colour—extra information, such as producing a determiner, was not scored although it was noted. For the other three puzzles, a point was given when participants produced the full DP including the determiner, the adjective(s) and the noun, as well as appropriate gender and syntactic structure. A second score was given for target feminine adjectives only (correct colour or size and gender) irrespective of the DP structure. A third qualitative analysis was performed on error types when the target DP was not produced. In these, only errors observed at least five times in each group were statistically analyzed.

Colour and size adjectives were analyzed separately because of their different properties, linguistic distributions, and age of acquisition. An important difference between these types is that colour adjectives are both variable and invariable in French, while size adjectives are always variable. Size adjectives are more frequent and are acquired slightly earlier, but are less varied, than colour adjectives (Royle & Valois, 2010). Colour adjectives are canonically post-nominal while size ones are pre-nominal (a typologically unusual position). Their different positions in the DP are considered to be linked to movement properties and features in the grammar of French (Valois, 1991).

Due to the small sample size, target and non-target responses were entered into non-parametric Kruskal-Wallis analyses with group as a between-subjects factor (DLD, AGE, MLU). Post hoc Mann-Whitney tests with Bonferroni corrections were used to directly compare groups when relevant. Analyses on effects of gender (masculine vs. feminine) or variability (variable vs. invariable) were run when adjectives in the puzzle bore these properties. Because comprehension levels were globally high in all children—the range in children with DLD was 96.9%–98.4%, depending on the puzzle—these data were not analyzed further.

Spontaneous speech. For each child, 200 utterances were examined to provide a reliable picture of his or her linguistic behaviour. Utterances were coded using the Systematic Analysis of Language Transcripts program (Miller & Chapman, 1984–2002; adapted for French by Elin T. Thordardottir, 2005). The first transcription was conducted by the research assistant who had administered the task. Each was checked by a second native speaker, then coded for morphosyntax and checked again. All disagreements were resolved by consensus or coded as incomprehensible (< 1%). The entire DLD corpus was re-transcribed by the first author. Inter-transcriber agreement was 96.6% for words and 97.9% for morphemes.

Between 71 and 171 DPs were produced by each child ($M = 127$, $SD = 29.7$), with a total of 3049 DPs included in the analysis. DPs containing unintelligible elements were excluded from the analysis (DPs per child: $M = 3.2$, $SD = 2.3$, Min. = 0, Max. = 10). Preposition use was considered when it occurred DP-initially in the form of complex contracted determiners (e.g., *du* and *de la* 'of-the.m/f'). Due to the small sample size and heterogeneous error distributions, Kruskal-Wallis analyses with group as a between-subjects factor (DLD, AGE, MLU) were used. Post hoc Mann-Whitney tests, with Bonferroni corrections for multiple comparisons, were applied to compare groups when relevant.

Results

Puzzles: Target Responses on Full Determiner Phrases and Variable Feminine Adjectives

Puzzles 1 and 2: Colour naming and size determiner phrases. Global results for target responses on each puzzle are reported in **Table 2**. Analyses reveal no differences among groups for both colour naming ($H = 3.79$, $p > .10$) and size DP production ($H = 1.16$, $p > .10$). As can be seen in **Table 2**, all groups showed high target response levels, with the most variability in the MLU group.

Analyses on target adjectives with feminine forms in size DPs (i.e., *grande/grosse*, 'big', or *petite*, 'small', $n = 4$) again revealed no group differences ($H = 0.8$, $p > .10$), with similar median and mean responses for the three groups. **Table 3** provides details on target feminine adjective responses for tasks 2 and 4.

Puzzle 3: Colour determiner phrases. Because two children with DLD did not complete both A and B versions of this puzzle, we tested whether both versions were equivalent in controls. A *t* test assuming unequal variance comparing the mean responses on version A ($M = 4.81$, $SD = 0.75$) and B ($M = 4.75$, $SD = 1.66$) of the control subgroups showed no significant differences, $t(14.39) = 0.12$, $p > .10$.

Following this, total target DP production scores were computed for 12 items by doubling the score on six items in order to include participants (DLD and controls) who did only task A. Analyses revealed a significant effect of group on target responses ($H = 8.74$, $p = .013$; see **Table 2**). Post hoc comparisons show that differences between the AGE and DLD groups reached significance with an adjusted alpha of .025 ($U = 52$, $z = 2.05$, $p = .020$, $d = 0.95$). No differences were found between the MLU and DLD groups, $U = 24$, $z = 0.79$, $p > .10$ (see **Table 2**). Because there were few feminine variable items in this puzzle (three if the participant did both A and B versions), we did not further analyze these items.

Puzzle 4: Size and colour determiner phrases.

Again, we checked whether both versions of the puzzle were equivalent. A *t* test assuming equal variance comparing controls' mean responses on version A ($M = 8.00$, $SD = 2.50$) and B ($M = 9.58$, $SD = 2.64$) showed no significant differences, $t(26) = 1.62$, $p > .10$.

Following this, total target production scores on 24 DPs were computed for analyses (see **Table 2**) by doubling the score on 12 items in order to include participants (DLD and controls) who only did task A. Analyses reveal a significant effect of group on target responses ($H = 7.27$, $p = .003$). Post hoc comparisons, with an adjusted alpha of .025, revealed a significant difference between the AGE and DLD groups ($U = 53$, $z = 2.15$, $p = .002$, $d = 1.22$), but not between MLU and DLD groups ($U = 0.21$, $z = 2.57$, $p > .10$). A comparison on feminine variable adjective targets ($n = 18$) transformed to a score over nine in order to include participants who did only task A, again revealed a significant effect of group on target responses ($H = 13.21$, $p = .001$; see **Table 3**). Post hoc tests revealed a significant difference between the DLD and AGE groups ($U = 61.5$, $z = 3.05$, $p = .001$, $d = 2.13$). The comparison between the DLD and MLU groups did not reach significance ($U = 46.5$, $z = 1.47$, $p = .07$).

Qualitative analyses of non-target responses on puzzles. In this section, we provide a detailed analysis of errors and non-target responses found in the elicitation task as they were expected to reveal different production strategies in the three groups. These responses were collated from the three puzzles involving DPs (puzzles 2, 3, and 4). The two children with DLD who did not perform both versions of puzzles 3 and 4 had their non-target responses on these tasks multiplied by two, while errors for matched-control pairs of children on A and B versions were added together to provide representative numbers of errors for a child who had done both versions. A total of 611 errors or non-target productions were observed across groups (DLD = 274, AGE = 87, MLU = 250). Only errors observed

Table 2
Target Responses for Colour Adjectives (Puzzle 1) and Full Determiner Phrases (Puzzles 2–4)

Task	<i>H, p</i>	<i>M</i>	Median	<i>SD</i>	Range
Puzzle 1 (colour naming, 6 items)					
DLD	3.79, .15	5.50	6	0.76	4–6
AGE		5.75	6	0.46	5–6
MLU		4.63	5	1.60	1–6
Puzzle 2 (size DPs, 8 items)					
DLD	1.16, .56	6.00	6	2.14	2–8
AGE		6.88	8	1.81	3–8
MLU		5.63	6.5	2.88	0–8
Puzzle 3 (colour DPs, 12 items, averaged over 6)^a					
DLD	8.74, < .05	4.31	4.75	1.41	1–5.5
AGE		5.31	5.5	0.46	4.5–6
MLU		4.19	4.5	0.96	2–5
Puzzle 4 (size and colour DPs, 24 items, averaged over 12)^a					
DLD	7.27, < .05	6.94	6.5	2.86	2–11
AGE		9.75	10	1.58	7–11.5
MLU		7.25	8	1.87	3.5–9.5

Note. AGE = controls matched on age; DLD = children with developmental language disorder; DP = determiner phrases; MLU = controls matched on mean length of utterance.

^aParticipants with DLD (*n* = 2) and controls (AGE, *n* = 2; MLU, *n* = 2) who only did version A of Puzzle 3 had the opportunity to produce 6 full DP structures, and for Puzzle 4 had the opportunity to produce 12 full DP structures.

Table 3
Target Response Results on Feminine Adjectives in Puzzles 2 and 4

Task	<i>H, p</i>	<i>M</i>	Median	<i>SD</i>	Range
Puzzle 2 (size adjectives, 4 items)					
DLD	0.8, .67	2.50	4	1.69	0–4
AGE		3.10	4	1.46	0–4
MLU		2.38	4	1.85	0–4
Puzzle 4 (colour and size adjectives, 18 items, averaged over 9)^a					
DLD	13.21, < .01	4.38	4.75	2.60	0–8
AGE		8.50	8.5	0.85	7.5–10 ^a
MLU		6.25	6.25	1.41	4.5–8

Note. AGE = controls matched on age; DLD = children with developmental language disorder; MLU = controls matched on mean length of utterance.

^aParticipants with DLD (*n* = 2) and controls (AGE, *n* = 2; MLU, *n* = 2) who only did version A of Puzzle 4 had the opportunity to produce 10 variable feminine adjectives.

more than five times in a given group were considered for the analyses. These were gender errors on size or colour adjectives (e.g., *La grenouille *vert* 'the.f frog green.m') or determiners (e.g., **La petit bateau blanc* 'the.f white.m small.m, a.m boat'), determiner omissions (e.g., *__ canard rouge* ' __ duck red'), and splitting the DP (e.g., *Le petit bateau blanc* 'the small white boat' → *Le blanc petit, un bateau* 'the.m white.m small.m, a.m boat').^{3,4}

Table 4 presents detailed information about observations. Kruskal-Wallis analyses with Group as a between-subjects factor (DLD, AGE, MLU) for gender errors on colour adjectives revealed a significant main effect of Group ($H = 10.39, p = .006$). Children with DLD

produced significantly more gender errors ($n = 62$) on colour adjectives than the AGE group ($n = 20; U = 4, z = 2.89, p = .002, d = 2.15$), but only showed a trend for a difference with the MLU group ($n = 43; U = 16, z = 1.63, p = .052, d = 0.83$). Of interest was that one child (D6) tended to overuse the non-default feminine form for green (*verte* [vɛ(ʁ)t]), which is observed nowhere else in the 151 control participants aged 3–6 years (unpublished data). A trend for an effect of Group was found for gender errors on size adjectives ($H = 5.14, p = .075$). Because children with DLD and the AGE group showed highly different patterns, we performed post hoc comparisons on these two groups. Differences were observed between DLD and AGE groups in the production of gender errors

	<i>H, p</i>	<i>M</i>	Median	<i>SD</i>	Range	Nb
Colour adjective gender						
DLD	10.39, <.01	7.75	8	2.76	3–12	8
AGE		2.50	2	2.07	0–6	6
MLU		5.38	5	2.97	1–11	8
Size adjective gender						
DLD	5.14, .078	5.75	5.5	5.57	0–14	5
AGE		0.63	0	1.41	0–4	1
MLU		5.38	5	3.80	0–10	6
Determiner gender						
DLD	0.10, .616	2.5	1	4.11	0–12	4
AGE		0.63	0.5	0.74	0–2	4
MLU		0.88	0	1.81	0–5	2
Determiner omission						
DLD	4.29, .117	6.25	0	12.24	0–35	3
AGE		0	0	0.00	0	0
MLU		7.13	2	11.15	0–32	5
Split DP						
DLD	1.22, .054	8.70	2	8.70	0–20	5
AGE		4.63	3	5.83	0–18	6
MLU		11.25	8	11.94	0–35	7

Note. AGE = controls matched on age; DLD = children with developmental language disorder; DP = determiner phrases; MLU = controls matched on mean length of utterance; Nb = number of children exhibiting this pattern, recall that control groups represent pairs of children.

³ Responses that were observed at least five times, but linked to fewer than three children in either group were word order errors (e.g., *Le bateau blanc* → *Le blanc bateau* 'the.m white boat'), and other errors (e.g., copula or conjoined DP structures: *Une maison rouge (elle) est petite* 'A red house (it) is small', or *Le bateau blanc et gros* 'The boat white and big').

⁴ Adjective commissions (e.g., *Purple* for *brown* or *mommy* for *big*) and determiner commissions (e.g., *La grenouille grise* → **Une grenouille *gris* 'A/one.f frog grey.m') were observed more than five times, but were rare in controls and thus not analyzed further.

on size adjectives, but these did not reach our alpha criterion of .025 ($n = 46$ vs. 5 ; $U = 15.5$, $z = 1.68$, $p = .047$, $d = 1.26$). No other differences between groups on error patterns were observed.

Spontaneous speech corpus. The children's spontaneous speech corpus was analyzed to compare similar DP structures across both tasks, but also other types not elicited with the puzzles. Based on 200 utterances per participant, we analyzed all DP contexts with determiners, adjectives, and other complex DP structures. All groups showed high levels of DP production, ranging from 74 in one MLU-matched child to 165 in one child with DLD⁵. Total numbers of DP structures produced by each group were 939 in the DLD group ($M = 130.8$, $SD = 71.6$), 1073 in the AGE group ($M = 134.1$, $SD = 20.2$), and 1046 in the MLU group ($M = 117.4$, $SD = 34.2$). A total of 302 analyzable errors were observed on DP structures. Children with DLD produced half of these (i.e., 155), AGE controls produced 63, and MLU controls 84. Four error types—agreement errors on adjectives or determiners, substitutions, omissions, and overregularizations—reached the five-item-per-group criterion. All other error types including word order errors, added elements, and other errors were negligible ($n \leq 5$ in a given group). Determiner and adjective agreement errors involved using an inappropriate gendered adjective or determiner in a determiner phrase (*la *vert* 'the.f green.m'). Substitutions involved definiteness errors (1a), number errors (1b), lexical-semantic errors (1c), and preposition substitutions (1d). Most omissions involved determiners but some involved nouns (e.g., *six* ___ 'six (years old)').

- (1) a. *des chevaux* for *les chevaux*
det.indef horse.pl det.def horse.pl
'some horses' 'the horses'
- b. *le traces* for *les traces*
the.m.sg [lœ] traces the.pl [lɛ] traces
'the traces' 'the traces'
- c. *un lion* for *un tigre*
'a lion' 'a tiger'
- d. *sa trompe dedans* for *la trompe sur*
l'épan [epã] *l'éléphant*
its trunk inside the.def.f trunk on
the-elephant the elephant
'his trunk inside' 'the trunk on
the elephant'

Four overregularization patterns were observed in the corpus. Three were linked to the non-application of obligatory morphophonological processes in contraction (2a), elision (2b), and liaison (2c). French-speaking children master contraction in elicitation by age 5 and some children at younger ages (approx. 3;01; Béchara, 2015). Overregularizations of contractions, elisions, or liaisons were rare but more common in participants with DLD. A fourth type of overregularization observed more frequently in controls was the misuse of irregular plural or singular nouns (2d). We checked all potential error types in the corpus outside the DP and only one other significant difference in patterns between groups was observed on verb number agreement, which was more common in children with DLD, but still quite rare (e.g., *Les fleurs *est belles*, 'the.pl flowers *is beautiful'; DLD: $M = 2.25$, $Mdn = 2$, Range = 2–8; AGE: $M = 0.38$, $Mdn = 0$, Range = 0–2; $z = -2.23$, $p < .05$).

- (2) a. *de les tirex* [dœletʰɪʁɛks] for *des tirex* [dɛtʰɪʁɛks]
'some t-rexes'
- b. *le nenfant* [lœnãfã] for *l'enfant* [lãfã]
'the child'
- c. *Lzami l'dinosaure* for *l'ami du dinosaure*
[lɛzãmidʰnozœʁ] [lamidyʰnozœʁ]
'the friend the dinosaur' 'the friend of the
dinosaur'
- d. *un chevaux* [ʃœvo] for *un cheval* [ʃœval]
'a horses' 'a horse'

We compared groups on the four identified error types with Kruskal-Wallis tests, and when relevant, post hoc Mann-Whitney tests with an adjusted alpha of .025. Descriptive information is provided in **Table 5**. Significant group effects were found for substitution ($H = 8.85$, $p = .012$) and omission errors only ($H = 10.58$, $p = .005$). Follow-up comparisons revealed significant differences in substitution errors produced by DLD and AGE groups ($U = 9.5$, $z = 2.31$, $p = .010$, $d = 1.56$), as well as with the MLU group ($U = 6.5$, $z = 2.63$, $p < .01$, $d = 1.77$). Omissions (e.g., *_ porte* '(the) door') were significantly more common in the DLD group than the AGE group ($U = 2.0$, $z = 3.10$, $p = .001$, $d = 1.65$), but not in the MLU group ($U = 16.5$, $z = 1.58$, $p = .057$, $d = 0.70$).

Based on the assumption that the narrow focus of the puzzles might have provided poorer information about

⁵ Note that numbers include proper noun phrases as these were occasionally introduced by the preposition *à* used in genitive structures as in *à Maman* 'Mommy's'. These usually accounted for less than 10% of the corpus.

Table 5**Descriptive Statistics for Error Productions on Determiner Phrases in the Spontaneous Speech Corpus**

	<i>H, p</i>	<i>M</i>	Median	<i>SD</i>	Range	Nb
Gender agreement errors						
DLD	3.32, .19	3.25	2.5	2.66	0–9	7
AGE		1.75	2	1.39	0–3	6
MLU		1.38	1.5	1.06	0–2	6
Omissions						
DLD	10.58, < .01	9.38	7	6.30	3–21	8
AGE		1.88	1.5	1.36	0–4	7
MLU		5.38	3.5	5.07	0–16	7
Substitutions						
DLD	8.85, .012	4.5	4.5	2.27	1–8	8
AGE		1.63	2	1.30	0–4	6
MLU		1.25	1	1.28	0–3	5
Overregularization						
DLD	0, 1	1.25	1	1.04	0–3	6
AGE		2.0	1	2.56	0–6	5
MLU		2.13	1	3.36	0–10	5

Note. AGE = controls matched on age; DLD = children with developmental language disorder; MLU = controls matched on mean length of utterance; Nb = number of children (out of 8) exhibiting this pattern.

adjective inventories and gender-marking abilities than the corpus, we reviewed each child's spontaneous production of these items and compared these to elicitation patterns (Table 6). Table 6 illustrates that only two children with DLD (D8 and D9) spontaneously produced four or more feminine variable adjectives; the other six did not exhibit productive use of these forms (Hiriarteborde, 1973). One (D8) showed normal scores on elicitation tasks, but another (D9) did not. The global inventory of feminine adjectives spontaneously produced by the DLD group was small: seven lemmas, four of which were tested in the tasks. We also noted that only D4 showed any signs of difficulties with gender marking on determiners in spontaneous speech. Most other errors observed on determiners were substitutions and omissions.

In sum, we observed a number of characteristics of DP production in two contexts: (a) a controlled barrier task with puzzles eliciting size and colour adjectives in simple and slightly more complex DPs and (b) in a spontaneous speech play context. The puzzle tasks revealed difficulties producing target configurations for DP structures involving

colour adjectives with little difficulties with size adjectives in children with DLD compared to age-matched controls, but few differences with MLU-matched controls. A finer analysis on error types revealed similar patterns on gender-marked colour adjectives: children with DLD produced more errors than age-matched controls, while trends for differences were observed on size adjectives. An analysis of corpus errors on similar DP structures revealed that gender errors were not a feature distinguishing groups, but rather that omission and substitution errors in the DP were salient features of spontaneous productions by children with DLD.

Discussion

Although our task was run on a small group of French-speaking children, some interesting and important patterns emerged. Our elicitation tasks revealed that globally, the DP is well mastered in these children. Any lexical-semantic errors on colour adjectives or in the corpus appear to cut across participant groups and thus do not appear to be the root cause for observed differences between them. Robust comprehension of these structures was also noted.

Table 6

Comparison Task Results for Adjective Production, and Adjective or Determiner Errors in Spontaneous Speech in Children with Developmental Language Disorder

	Elicitation			Corpus analysis					
	Tasks (target responses)			Adjective use			Determiner		
	2	3	4	Variable		Total adjectives	Errors ^c		
	Size	Col.	Siz. + Col.	Masc.	Fem.	Lemmas	Lexemes (Lemmas)	Masc.	Fem.
1. D2	2/8	4/6	4/12	3/3	2/5	4	16 (8)	3 (1)	6 (1)
2. D3*	6/8	9/12	12/24	3/3	2/2	4	6 (5)	4 (0)	0 (0)
3. D4*	6/8	1/6	2/12	9/9	0/3	4	13 (7)	16 (3)	9 (4)
4. D5	6/8	10/12	12/24	17/17	1/1	6	29 (12)	7 (0)	4 (2)
5. D6	4/8	10/12	14/24	29/29	2/3	7	38 (11)	12 (0)	7 (0)
6. D7*	8/8	9/12	18/24	34/35	2/2	10	54 (32)	4 (1)	1 (0)
7. D8	8/8	11/12	21/24	11/12	3/4	5	39 (21)	2 (1)	1 (0)
8. D9	8/8	10/12	19/24	12/12	8/8	10	36 (20)	2 (0)	5 (0)
Mean	5.6/8	8.6/12 ^a	14/24 ^b	0.98	0.75	6.25	29 (14)	6.25	2.14
SD	1.61	2.83	6.35			2.55	15.96 (9.2)	5.15	3.23
Median	6	9.5	14			5.50	32.50 (11.5)	4	4.5

Note. Results are presented as number target/total number or number of different lemmas. Bold numbers indicate that scores on the elicitation tasks are within normal range, as defined by scores for the age-matched controls. Fem. = feminine; Lemma = number of different words produced; Masc. = masculine; Total = Nb of items produced.

Target/total productions: ^aaveraged over 12 items; ^baveraged over 24 items; ^cIncludes omission, agreement and substitution (e.g., definiteness), errors specific to gender are in parentheses. * = indicates children who made determiner errors in the elicitation task.

However, specific difficulties were found when children with DLD were asked to produce DPs with variable adjectives, mostly feminine colour adjectives. Determiner errors were only salient in some children: D4 omitted a quarter of his determiners and made one definiteness error, and D7 omitted two and made six gender errors on them, most often using feminine for the masculine, which is highly unusual, even in clinical observations. Finally, D3 omitted or substituted a smaller number of determiners. Children with DLD were also more likely to produce many more errors than age-matched controls, especially on adjectives, whether size or colour. Note that these size adjectives are acquired at age 3 in French (Royle & Valois, 2010; Valois & Royle, 2009; Valois, et al., 2009).

Globally, our task results are consistent with cross-linguistic data from Swedish and Russian, as well as previous

French studies of DP production. As mentioned above, older Russian children with DLD have difficulties producing gender-marked adjectives in an antonym elicitation task (e.g., *open* vs. *closed*; Tribushinina & Dubinkina, 2012). Gender difficulties on Swedish adjectives in elicited DP probes also have been observed, but were less salient than determiner-omission and substitution errors (Leonard et al., 2001).

Our results on adjective production, however, are not as clear-cut as those provided in Roulet-Amiot and Jakubowicz (2006) who showed important difficulties with variable adjectives in children with DLD. This could be explained by their use of non-contrasting pictures where adjectives were not pragmatically salient. It is also possible that some of their items were much harder than those in our task. For example, they used derived adjectives and did not strictly control for age-of-acquisition norms.

Therefore, we cannot be sure the task was not tapping into lexical-semantic knowledge. Regarding determiner errors, in the sentence elicitation task reported above, Pizzioli and Schelstraete (2008) observed determiner omissions in older French-speaking children with DLD. However, the tasks they used were quite different from ours.

We checked whether a spontaneous speech corpus would be more instructive in identifying agreement or DP-structure difficulties in children with DLD. Spontaneous speech analyses highlighted other differences between French-speaking children with DLD and their peers, especially omission and substitution errors, which were more common in the DLD group than both control groups. Although agreement errors are found on determiners and adjectives, they were relatively rare in our study, but adjectives were also under-represented in the corpus. In particular, and as found with younger children, the corpus provides little information about adjective agreement abilities, and might even over-evaluate them if, for example, we were to score total correct adjective production or even masculine variable adjective production (Table 6, columns 5 and 6). Our data were not coherent with previous studies of determiner production in spontaneous speech showing little difficulties in determiner production (e.g., low rates of omission in obligatory contexts) in French-speaking children with DLD (Le Normand et al., 1993; Paradis & Crago, 2001). One reason for this disparity may be that in Le Normand et al.'s (1993) study, only 29 DP contexts on average were analyzed per child. The Paradis and Crago (2001) study, however, does not appear to have this issue as a 45-minute sample of spontaneous speech was taken, which was similar in length to our sample.

Contrasting elicitation and spontaneous-speech samples, we can affirm that both approaches were useful in evaluating adjective agreement, DP structure, and lexical-semantic abilities, and also revealed significant difficulties with determiners in children with DLD. Only the puzzle tasks revealed gender agreement as an area of weakness in our participants. Because French adjective morphology is opaque (Royle, 2011) and thus difficult for all children, differences between typical and impaired language development were not as robust as what has been found for other languages with productive and transparent morphology. However, we did observe particular patterns only in children with DLD, namely overuse of feminine for masculine forms on determiners and adjectives, which is highly unusual. This behaviour could be a clear marker of language disorders in French, and should be investigated further. Royle and Stine (2013) suggested that some errors observed in the corpus are worth pursuing in further

elicitation studies, although they do not stand out in the spontaneous data. These include morphophonological alternations in elision, liaison, and contraction (see examples in (1) above), which are easily elicited in controlled contexts (Béchara, 2015).

Regarding other errors found in the corpus, only verb number agreement errors stand out as a potential marker for DLD in these children, and these are quite rare. It is well known that recording and coding a spontaneous speech corpus demands more time and effort to implement than a short elicitation task. Thus, although some interesting features of DLD speech can be extracted from the corpus, we believe elicitation tasks hold the potential to rapidly identify children with language production difficulties in an ecological way. While not discounting the usefulness of spontaneous speech samples for providing global measures of lexical richness, syntactic and morphosyntactic abilities, we acknowledge the puzzles' utility to quickly tap into agreement and short sentence structure difficulties in French children, and their potential to help identify children with language development delay or disorders in their pre-school years. It is also relevant from a clinical point of view, as well as from developmental and theoretical points of view, to distinguish colour and size adjectives as we have done, when evaluating DP production in children with DLD. Remember that we observed that children with DLD did not distinguish themselves from controls on size adjectives, which are acquired early, but showed worse results on colour ones. Grouping these two types of adjectives in a speech corpus analysis or in an elicitation task could water down interesting information about a child's linguistic abilities or would make it more ambiguous where the difficulties were observed.

Steel et al. (2013) emphasized that spontaneous speech data often underestimate children's linguistic competence, but as we have shown, it can also overestimate it because children may opt to use structures they master (Leonard, 2016). Two previous studies of Spanish-speaking children showed that elicitation revealed that those with DLD had difficulties producing plural nouns, while spontaneous speech did not distinguish them from typically developing children (Bedore & Leonard, 2001, 2005). In our study, difficulties with determiners varied according to task. Table 6 highlights the fact that only D3, D4, and D7 had difficulty with determiners on the puzzles, but most participants with DLD showed determiner errors (i.e., agreement, substitution, or omission) in spontaneous speech. The opposite held for adjective agreement abilities. All participants with DLD, except D8, showed difficulties on

puzzle 4, and all produced gender-marking errors on the tasks but only half (i.e., D2, D4, D6, and D8) showed any errors (between one and three) on the small number of feminine adjectives they use in the corpus.

Additionally, spontaneous speech samples often do not provide sufficient samples for contexts that the clinician or researcher wants to investigate (Gerken, 2000). This is definitely the case for gender-marked adjectives and complex DPs, which are rare in the corpus. This could be due to pragmatic factors (e.g., the referent is salient, no extra information is needed to identify it) or to avoidance strategies, and cannot be clarified in this type of data. Thus, we believe it is useful to apply multiple methods to build a complete picture of the child's linguistic abilities. In the case of D8, none of the measures, except error rates on the task, seemed to highlight linguistic difficulties. However, rapid identification of specific difficulties might be better served by elicitation tasks. Eisenbeiss (2011) highlighted the tension between using elicitation experiments, which can be challenging for children, and obtaining insufficient information from spontaneous speech. However, the first three puzzles used here can be used with typically developing French 3-year-olds without difficulty (Royle & Valois, 2010) and were developed to ecologically reflect other puzzles available to parents or daycares. They thus hold the potential to provide information on lexical, syntactic, and agreement abilities in young French-speaking children.

Limitations

A limitation of this study was the small sample size for the group with DLD. Data from larger samples are required to confirm the usefulness of these puzzles as a screen for oral language disorders in children aged 5 to 6 years. Furthermore, two of the children were not presented with the B versions of puzzles 3 and 4. Both A and B versions should be used for the same child for clear norms to emerge. It is possible that the easiest puzzles could be useful in identifying younger children with DLD (e.g., between ages 3 and 5), but this has not yet been established. Also, these tasks might prove difficult for second language learners who notoriously find French gender agreement difficult, even if their mother tongue has this feature (see Royle, Bergeron, & Marquis, 2015). Thus, this task would not necessarily be useful in identifying language impairment in contexts where a learner has not yet fully mastered French. Studies on second language mastery of gender agreement would be useful to further deepen our understanding of these issues.

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Appendix A

Score-Sheets With Stimuli for Version A

Table 1A		
Score-Sheet with Stimuli for the Version A of Task 1		
Item	Colour	Production
Pre-test 1	<i>rose</i> 'pink'	
Pre-test 2	<i>bleu</i> 'blue'	
Test 1	<i>jaune</i> 'yellow'	
Test 2	<i>noir</i> 'black'	
Test 3	<i>rouge</i> 'red'	
Test 4	<i>vert</i> 'green'	
Test 5	<i>brun</i> 'brown'	
Test 6	<i>blanc</i> 'white'	

Table 2A			
Score-Sheet with Stimuli for the Version A of Task 2			
Item	Object	Size	Production
Pre-test 1	<i>cheval</i> 'horse'	<i>gros</i> 'big'	<i>Le gros/grand cheval</i> 'the big horse'
Pre-test 2		<i>petite</i> 'small'	<i>Le petit cheval</i> 'the small horse'
Test 1	<i>bateau</i> 'boat'	<i>gros</i> 'big'	<i>Le gros/grand bateau</i> 'the big boat'
Test 2		<i>petite</i> 'small'	<i>Le petit bateau</i> 'the small boat'
Test 3	<i>canard</i> 'duck'	<i>gros</i> 'big'	<i>Le gros/grand canard</i> 'the big duck'
Test 4		<i>petite</i> 'small'	<i>Le petit canard</i> 'the small duck'
Test 5	<i>maison</i> 'house'	<i>gros</i> 'big'	<i>La grosse/grande maison</i> 'the big house'
Test 6		<i>petite</i> 'small'	<i>La petite maison</i> 'the small house'
Test 7	<i>grenouille</i> 'frog'	<i>gros</i> 'big'	<i>La grosse/grande grenouille</i> 'the big frog'
Test 8		<i>petite</i> 'small'	<i>La petite grenouille</i> 'the small frog'

Table 3A
Score-Sheet with Stimuli for the Version A of Task 3

Item	Animal	Colour	Production
Pre-test 1	<i>cheval</i> 'horse'	<i>rose</i> 'pink'	<i>Le cheval rose</i> 'the pink horse'
Pre-test 2		<i>bleu</i> 'blue'	<i>Le cheval bleu</i> 'the blue horse'
Test 1	<i>canard</i> 'duck'	<i>jaune</i> 'yellow'	<i>Le canard jaune</i> 'the yellow duck'
Test 2		<i>noir</i> 'black'	<i>Le canard noir</i> 'the black duck'
Test 3		<i>blanc</i> 'white'	<i>Le canard blanc</i> 'the white duck'
Test 4	<i>grenouille</i> 'frog'	<i>rouge</i> 'red'	<i>La grenouille rouge</i> 'the red frog'
Test 5		<i>verte</i> 'green'	<i>La grenouille verte</i> 'the green frog'
Test 6		<i>brune</i> 'brown'	<i>La grenouille brune</i> 'the brown frog'

Note. To combine versions A and B, one can present the same animals with all six colours.

Table 4A

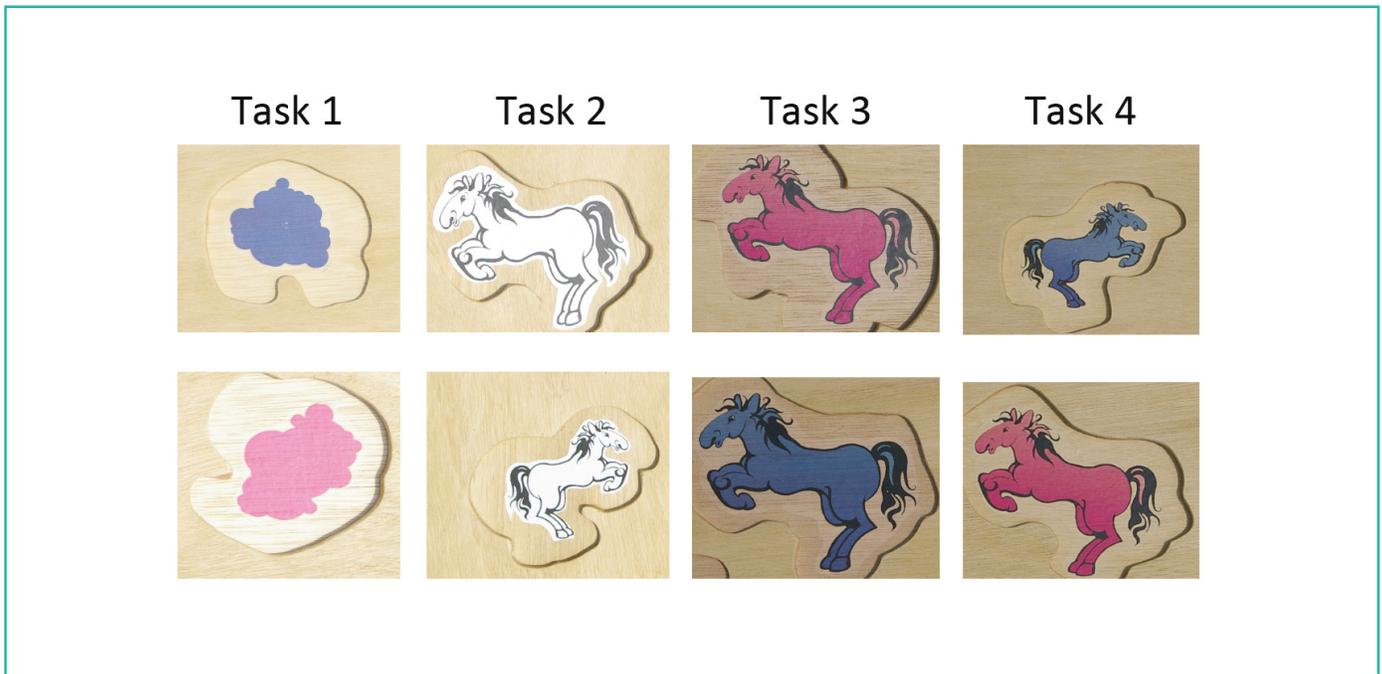
Score-Sheet with Stimuli for the Version A of Task 4

Item	Object	Size	Colour	Production
Pre-test 1	cheval 'horse'	<i>petite</i> 'small'	<i>bleu</i> 'blue'	<i>Le petit cheval bleu</i> 'the small blue horse'
Pre-test 2		<i>grosse</i> 'big'	<i>rose</i> 'pink'	<i>Le gros/grand cheval rose</i> 'the big pink horse'
Test 1	bateau 'boat'	<i>grosse</i> 'big'	<i>jaune</i> 'yellow'	<i>Le gros/grand bateau jaune</i> 'the big yellow boat'
Test 2			<i>noir</i> 'black'	<i>Le gros/grand bateau noir</i> 'the big black boat'
Test 3			<i>blanc</i> 'white'	<i>Le gros/grand bateau blanc</i> 'the big white boat'
Test 4		<i>petite</i> 'small'	<i>jaune</i> 'yellow'	<i>Le petit bateau jaune</i> 'the small yellow boat'
Test 5			<i>noir</i> 'black'	<i>Le petit bateau noir</i> 'the small black boat'
Test 6			<i>blanc</i> 'white'	<i>Le petit bateau blanc</i> 'the small white boat'
Test 7	maison 'house'	<i>grosse</i> 'big'	<i>rouge</i> 'red'	<i>La grosse/grande maison rouge</i> 'the big red house'
Test 8			<i>verte</i> 'green'	<i>La grosse/grande maison verte</i> 'the big green house'
Test 9			<i>brune</i> 'brown'	<i>La grosse/grande maison brune</i> 'the big brown house'
Test 10		<i>petite</i> 'small'	<i>rouge</i> 'red'	<i>La petite maison rouge</i> 'the small red house'
Test 11			<i>verte</i> 'green'	<i>La petite maison verte</i> 'the small green house'
Test 12			<i>brune</i> 'brown'	<i>La petite maison brune</i> 'the small brown house'

Note. To combine versions A and B, one can present the same objects with all six colours.

Appendix B

Sample of Images Used for the Tasks





Barriers and Facilitators to Cultural Competence in Early Hearing Loss Services: A Qualitative Analysis



Obstacles et facilitateurs à la compétence culturelle dans les services précoces offerts aux enfants ayant un trouble auditif : une analyse qualitative

KEYWORDS

CULTURAL COMPETENCE

EARLY HEARING
DETECTION AND
INTERVENTION

QUALITATIVE INQUIRY

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Abstract

Increasing diversity in Canada has direct implications for early hearing detection and intervention. Efforts to improve cultural competence in early hearing detection and intervention should be informed by evidence on how cultural differences can affect services; however, there is limited empirical research in this area. The objective of this study, therefore, was to explore the experiences of practitioners in pediatric hearing loss services in providing care to families of minority culture backgrounds. To address this objective, a qualitative research design with semi-structured interviews was used to gain insight into practitioner perceptions of barriers and facilitators to the provision of culturally competent care. A total of 19 practitioners participated in this study. Three themes emerged from the interview data: characteristics of a culturally competent practitioner, barriers to service provision, and facilitators to service provision. Practitioners encountered barriers throughout the process of service delivery with language barriers affecting every stage. Practitioners also reported using various facilitators, such as communication strategies, to mitigate many of these challenges. This study contributes insight to a field that has received little attention in early hearing detection and intervention.

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Abrégé

L'accroissement de la diversité au Canada a des répercussions directes sur les services de détection et d'intervention précoces des troubles auditifs. Les efforts visant à améliorer la compétence culturelle des praticiens fournissant des services de dépistage et d'intervention précoces des troubles auditifs devraient être guidés par des données probantes qui indiquent comment les différences culturelles peuvent affecter les services offerts. Or, les recherches empiriques dans ce domaine sont limitées. L'objectif de cette étude était donc d'explorer l'expérience de praticiens travaillant auprès d'une clientèle pédiatrique ayant un trouble auditif et offrant des soins à des familles issues des cultures minoritaires. Un devis de recherche qualitatif et utilisant des entrevues semi-structurées a été sélectionné pour mieux comprendre les perceptions des praticiens quant aux obstacles et aux facilitateurs à la prestation de soins culturellement adaptés. Au total, 19 praticiens ont participé à cette étude. Trois thèmes sont ressortis des données des entrevues : les caractéristiques d'un praticien culturellement compétent, les obstacles à la prestation de services et les facilitateurs à la prestation de services. Les praticiens ont rencontré des obstacles tout au long du processus de prestation de services, la barrière linguistique affectant chaque étape. Les praticiens ont également indiqué avoir eu recours à divers facilitateurs, tels que des stratégies de communication, pour pallier à plusieurs de ces difficultés. Cette étude contribue aux connaissances d'un domaine des services de détection et d'intervention précoces des troubles auditifs qui a reçu peu d'attention.

Increasing diversity in Canada has direct implications for pediatric hearing loss services. The latest census projects that by 2036 almost half of Canada's population will be immigrants or children of immigrants (Statistics Canada, 2017). In such a multicultural population, cultural differences can create challenges for health care systems, which are typically tailored to meet the needs of the majority population. Culture can influence values, beliefs, and health-related practices, and can impact all aspects of health care service delivery (Betancourt, Green, Carrillo, & Ananeh-Firempong, 2003).

Consideration of culture is of importance in pediatric hearing loss services as culture can influence how families respond to services at various stages in the process, from identification to intervention. For example, hearing loss is stigmatized in some cultures and can be seen as the fault of the parents and a source of shame (Jackson, Traub, & Turnbull, 2008). Having a child with permanent hearing loss can even result in family relationships being severed (Jackson et al., 2008; Yucel, Derim, & Celik, 2008). In some situations, parents may refuse interventions to conceal their child's hearing loss. In addition, concerns have been raised about the additional stress multicultural families may experience when their cultural norms differ from those of practitioners and the interventions they use (Phillips, Worley, & Rhoades, 2010). Thus, practitioners who provide early hearing detection and intervention (EHDI) services should be aware of cultural differences and practice culturally competent care.

In their literature review, Betancourt et al. (2003) defined cultural competence in health care as

...understanding the importance of social and cultural influences on patients' health beliefs and behaviors; considering how these factors interact at multiple levels of the health care delivery system; and, finally, devising interventions that take these issues into account to assure quality health care delivery to diverse patient populations. (p. 293)

Efforts to improve cultural competence in EHDI programs should be informed by evidence on how cultural differences can impact services; however, our scoping review identified limited empirical research in this area (Grandpierre et al., 2018). In addition, available literature is dated, anecdotal, and possibly reflects stereotypes (Ball & Lewis, 2014; Eriks-Brophy, 2014). Although there is extensive research on cultural competence in other rehabilitation fields which detail the perspectives of practitioners and patients of minority culture backgrounds (i.e., Al Busaidy & Borthwick, 2012; Centeno, 2009; Dogan, Tschudin, Hot, &

Özkan, 2009; Dressler & Pils, 2009; Drolet et al., 2014; King, Desmarais, Lindsay, Piérart, & Tétreault, 2015; Lindsay, King, Klassen, Esses, & Stachel, 2012; Yang, Shek, Tsunaka, & Lim, 2006), one source of untapped information is the viewpoint of practitioners in pediatric hearing loss with respect to their experiences providing services to cultural minorities. Therefore, the goal of this study is to contribute empirical data to address this research gap.

Context

Most provinces in Canada have implemented Newborn Hearing Screening services. In Ontario, the main context of the study, each infant is screened at birth for hearing loss. If a referral is required, infants will then receive diagnostic assessments. The confirmation of a hearing loss diagnosis will lead to intervention discussions with audiologists regarding technological options as well as language therapy options. Language therapy options typically include spoken language approaches (oral), visual approaches (sign), and total communication approaches (oral + sign). In the context of this study, auditory verbal therapists who participated reported to prescribe to spoken language approaches. All services are publicly funded, excluding the purchase of hearing technology.

Objectives

As part of a larger program of work that investigates cultural influences on EHDI programs from the perspectives of both practitioners and families of minority culture backgrounds, the objective of this study was to explore the experiences of practitioners in the provision of services to families of minority culture backgrounds.

Method

Participants

Practitioners in the field of childhood hearing loss (e.g., audiologists, listening and spoken language therapists, itinerant teachers of the deaf and hard of hearing, speech-language pathologists) were eligible to participate if they provided (a) early hearing detection and intervention services to children with permanent hearing loss and (b) services to families of minority culture backgrounds (e.g., not the dominant culture in Canada: English and/or French Canadian).

Health care practitioners offering services to children with permanent hearing loss were recruited from the Children's Hospital of Eastern Ontario (CHEO), a tertiary care hospital located in Ottawa. The Professional Practice Leader of Audiology in CHEO's Audiology Clinic was approached regarding recruiting her team for the study.

Upon approval, all team members were approached for recruitment. Recruitment also occurred at a local conference in Ottawa (i.e., the Dual-Language Learning conference held in 2017), which was open to practitioners in the fields of audiology and speech language pathology. During scheduled breaks, practitioners were approached about their interest in participating.

Recruitment and data collection was informed by Thorne, Kirkham, and MacDonald-Emes's (1997) Interpretive Description method. Interpretive Description draws strongly on features of grounded theory, ethnography, and naturalistic inquiry and attempts to gather a meaningful account of a clinical phenomenon of interest and make it accessible to clinical understanding. This can be achieved by drawing on data collected from small samples through methods such as interviews. A convenience sample of 12–20 participants was therefore anticipated to be suitable for obtaining relevant information. This study received ethical clearance from the research ethics boards of Children's Hospital of Eastern Ontario Research Institute (16/01X) and the University of Ottawa (A10-16-01).

Procedure

Data were collected through individual interviews conducted in English. Prior to the interviews, participants were asked to complete a brief form to record demographics and job-related information. This included data such as profession title, practice setting, years of experience, and percentage of time working with patients of minority culture backgrounds. A semi-structured format was used to guide the interview from a pre-determined list of questions developed by the lead researcher (see Appendix). The interview protocol was informed by findings in our scoping review (Grandpierre et al., 2018). The interview consisted of open-ended questions on cultural sensitivity training (if received, from where, etc.), and general experiences with service provision to families of minority culture backgrounds. Prompts to seek new leads or request clarifications were incorporated into the interview protocol. Seven interviews were conducted over the phone due to distance (e.g., five were located outside Ottawa, two participants were not available to be in Ottawa at the time of the interview). Live interviews took place at practitioners' offices in CHEO. Field notes were taken by the researcher (VG) during interviews. All interviews were conducted by the researcher (VG).

Analysis

Interviews were audio recorded and transcribed verbatim. In addition, field notes were recorded and consulted during analysis. All data were entered into NVivo

(version 10.1.2), a qualitative software program used for coding and qualitative research analysis. Demographic information was entered into Excel.

Analysis of transcripts occurred concurrently with data collection (DiCicco-Bloom & Crabtree 2006). In the Interpretive Description method, inductive data analysis techniques are often used to highlight thematic patterns and commonalities to help characterize the topic of interest (Thorne et al., 1997). Inductive techniques are typically used in qualitative methods where data, such as transcripts, are analyzed to generate ideas (Thorne, 2000). Transcripts were analyzed using a coding process known as the *constant comparative method* based on Strauss and Corbin's (1990) open, axial, and selective coding methods, an approach that fits well within the Interpretive Description methodology (Thorne, Kirkham, & O'Flynn-Magee, 2004). Open coding involves reviewing and assigning labels to each passage. Axial coding involves a comparison of characteristics for each label among interviews. In selective coding, concepts become further refined by examining similarities of labels and collapsing these categories into major themes.

Abiding by qualitative research practices, the concept of trustworthiness (Krefting, 1991) was used to ensure quality and transparency in this study. Components of trustworthiness are credibility, transferability, dependability, and confirmability. Credibility was evidenced by data collected from various participants in different fields within audiology. Transferability was demonstrated by the collection of in-depth data with detailed descriptions of the setting and the participants. Dependability was achieved with a clear description of the research process. Reflexivity is an additional component to ensuring trustworthiness which enables transparency (Korstjens & Moser, 2018). In this study, reflexivity was achieved by the researcher documenting reflexive notes during the interviews, as well as the interview setting. In addition, peer debriefing with the coauthors occurred throughout each stage of the study. Finally, confirmability was evidenced using detailed field notes to help ensure neutrality of the data. Consulting coauthors with expertise in qualitative research, health sciences, and medical research about the decision-making on the research process also contributed to achieving confirmability.

Results

Participant Characteristics

A total of 26 practitioners responded to the invitation to participate; the study protocol was reviewed for feasibility.

As this topic is underexplored, all 26 respondents were elected to be interviewed; however, only 19 participated. Of those, 14 practitioners were recruited at CHEO and five from a local conference; the latter group practiced at centres (e.g., public and private health care facilities) in various cities in two Canadian provinces (Ontario and Nova Scotia). One participant provided written feedback to the interview questions but did not participate in an oral interview. **Table 1** provides a description of participant characteristics.

To gain multiple perspectives from practitioners in different roles within pediatric audiology, a diverse sample

of various professions was selected. Practitioners were audiologists, auditory-verbal therapists, an auditory-verbal educator, itinerant teachers of the deaf and hard of hearing, a hearing resource teacher, a case assistant worker in audiology, and speech language pathologists who work in audiology clinics. Experience in these fields ranged from 3 to 40 years.

Most of the practitioners had reported receiving prior education on cultural sensitivity in the form of training and/or lectures, varying from 1-hour lectures to a series of workshops. The majority were French and/or English speakers with Canadian and/or Francophone (Canada,

Table 1	
Participant Characteristics	
	All practitioners
Number (%)	19 ^a (100)
Sex, n (%)	
Male	1 (5.2)
Female	18 (94.7)
Languages^b, n (%)	
English	19 (100)
French	16 (84.2)
American Sign Language	2 (10.5)
Arabic	1 (5.2)
Spanish	1 (5.2)
Cultural backgrounds^b, n (%)	
European	10 (52.6)
Francophone (Canada, France)	7 (36.8)
Canadian	7 (36.8)
Asian	1 (5.2)
Australian	1 (5.2)
Position, n (%)	
Audiologist	7 (36.8)
Speech language pathologist	3 (15.7)
Therapists/teachers/case worker	9 (47.3)
Years of experience, median, (range)	17 (3–40)
Received cultural sensitivity education, n (%)	12 (63.1)

Note. ^aOne participant provided a written response to the interview protocol (e.g., was not available for an oral interview); ^bMany practitioners were multilingual and multicultural.

France) cultural backgrounds. Practitioners serviced a large variety of cultures, mainly consisting of Canadian Indigenous, Asian, and African.

The purpose of the interviews was to explore practitioners' experiences providing services to families of minority culture backgrounds. Three themes emerged from the interview data: barriers to service provision, facilitators to service provision, and characteristics of a culturally competent practitioner. It is important to note that most of the practitioners (74%) commented on the difficulty of attributing their patients' families' responses to cultural differences as opposed to typical stress responses. The results, therefore, represent when practitioners felt confident that their experiences with families reflected cultural differences.

Barriers to Service Provision

Practitioners described various barriers to care. Barriers were encountered throughout the care process, such as during audiological testing, diagnosis, amplification, language assessments, and interventions.

Language barriers. Language barriers affected every stage of treatment and were noted to be problematic even when professional interpreters or someone who could translate (e.g., colleagues or extended family members) were available: "Language barriers can be the biggest impediment for the family I think. In terms of making sure...let's say, often...we have one parent who speaks English and...they are acting as the interpreter for their husband which really is not ideal" (Participant 22). Another participant noted,

I think the hardest thing when working with families is when French or English is not their first language and they are working with an interpreter. What happens is that when an interpreter is working with you, you don't know how much filtering is going on because you don't know that other language, so when you are working with a family to get informed consent for something like a CI [cochlear implant] surgery you really want to make sure that parent really understands and certainly we've had families with English as a second language where you're hoping that the parent is saying that this is what they want and you are trying to make sure they have all the tools needed to make the right choice, with that interpreter. (Participant 7)

Language barriers also occurred in service provision when parents' interaction with their children at home was minimal. The level of parent-child interaction varies with culture and can affect services (e.g., spoken language

development) that depend on family engagement for optimal outcomes. For example, auditory verbal therapists typically promote ongoing verbal communication in families to support language development; however, if there is minimal parent-child interaction in a culture, this can reduce the effectiveness of the intervention:

So how do the parents interact with the kids because... they may not talk to their child as much or interact in the same ways we might expect so I think there has to be some sensitivity around what our expectations are for interacting with their kids...there's not only the nonverbal but the verbal.... In terms of nonverbal, I've worked with - it was actually the Indigenous population in Canada where eye contact was very different. So they don't give direct eye contact when they communicate. So being aware of that was really important as a clinician. (Participant 10)

Cultural challenges during audiological testing. In addition to language barriers, challenges to audiological testing were noted when caregivers were not comfortable receiving services because of the practitioners' gender (e.g., families sometimes preferred male practitioners). Practitioners described instances where families were not receptive to the information provided. Sometimes male practitioners were requested to relay the information. Other situations were more delicate, such as requesting the removal of children's religious attire during audiological testing.

When a child is coming in for a hearing assessment and we have a 30 minute time and from a clinical perspective it would be very lightly touched on if a child came in and for example, they wear a hijab and you had to ask them to remove it and there was a male audiologist at the time, he would be asked to change [to leave] at that time because that would be appropriate. (Participant 7)

Cultural challenges during discussions of hearing loss diagnosis. Almost all practitioners commented on surprising reactions when diagnoses were communicated, which were attributed to cultural differences. They discussed how, in some cultures, disability is stigmatized and seen as something shameful. This perception sometimes led to caregivers denying the hearing loss or refusing amplification for their children.

...when I worked in [Canadian province], I worked with many Asian families. And it was seen within...at least the family group of one of the patients that I worked with... the family was embarrassed about the diagnosis and so it was very challenging to get them to accept that

the hearing loss was in fact a permanent thing and to get them to accept that if they wanted a listening and spoken language mainstream-schooling outcome, the way to achieve that was through regular hearing aid use and therapy. One family in particular, they sought out alternative medicine to try and cure the hearing loss. (Participant 10)

I have experienced two middle eastern families where the parent made it very clear, that when they went back to [home country] to visit, that they took the hearing aids off. And the mother said "I don't even wear my glasses when I go back to Lebanon. I wear glasses now because I need them - but if I wear glasses as a young woman, I never would have found a husband." (Participant 12)

Cultural challenges during administration of standardized language assessments. Standardized language assessments also presented challenges for practitioners. Specifically, the assessment content was not always reflective of everyday environments. Most of the practitioners commented on how these assessments were normed on majority populations and that culturally inappropriate items affected scoring.

...having the proper assessment tools, [it is] very important. For example, you cannot use - immigrant families coming to Canada, [they have never seen] the Christmas tree and they never see snow, and they don't have any [idea of] what the snow looks like, they [have] never seen it in their country, so it would be like "what".... It's not appropriate to talk about snow without them experiencing it.... I cannot assess them with snow. Like I'm talking to a Canadian child about the desert and camel, they [have] never seen a camel and I cannot mention a camel in my first assessment because they don't have any experience with the camel, what it looks like, what sounds it makes, you know? So this knowledge is very important. (Participant 15)

Cultural challenges during language interventions. Many practitioners commented on barriers encountered when providing interventions to families of minority culture backgrounds. A variety of reasons were indicated, such as language barriers, limited culturally sensitive materials, differences in expectations between practitioner and parents regarding who will do the therapy, and differences in language output expectations between practitioner and parents: "I have to say, some of our books are white-centric, but I have newer books that I've bought for the little ones that have more different racial groups represented" (Participant 13). Another participant noted,

In some families...a few African families - no toys at home. And so we had to either provide some toys to show the mom what we wanted to do with them and modify our expectations in terms of sort of seeing what the mom did do with the kids and maybe building routines around or building language into daily routines and you know, dressing, and having the child help you know with food, meal time and that kind of thing. Um so ...I guess the cultural expectation of what a parent does in terms of how much do they actually play with their child or talk to their child - that would come into play. (Participant 12)

Facilitators to Service Provision

Although many challenges were noted, practitioners proposed a number of strategies to overcome barriers. Facilitators included communication strategies, learning about cultural differences, and strategies throughout the process of intervention.

Communication strategies. Communication strategies included ensuring comprehension by asking caregivers to repeat what was said. Using simple language, learning key phrases in the caregivers' language, speaking slowly, and using visual aids (e.g., writing key words or showing pictures or videos) were also seen as useful.

When we ask people if they understand what we are saying, and we know they have a different first language, initially when people nodded I assumed they were getting along ok. I learned over time that this is good in counselling but this is particularly important when there is a language barrier is that you need to get them to repeat back to you what they think you've said. Sometimes you see that there are huge gaps. (Participant 5)

I find written communication as a backup, even if I'm there in person, so I always have "what we do," "what we did" in writing so that they can go [to] somebody and say what exactly does this mean or, those kinds of things. So I would certainly use simpler speech, vocabulary, or I would show them what I'm talking about through either a picture or I would Google it or I would show them physically something in their home. I would say in English, you know, what do you refer this for example a sofa or couch, what would you call that? (Participant 19)

Strategies to learn about cultures. Many practitioners stated that learning about cultural practices was an important facilitator to culturally competent service provision. Strategies to learn about cultural differences included attending lectures, workshops, or training and

asking families questions about their daily routine, religious holidays, cultural traditions, and practices. Some stated that asking families about how disability was perceived provided helpful feedback on how to approach interventions. In addition, home visits were considered invaluable for learning about differences to better tailor care to meet the needs of the family.

If they've come from a country where this is viewed as a stigma then the first question to ask is how is hearing loss viewed in your culture...I try to get the info from the family, how do you feel about this. Then I can address their concerns, will he struggle wearing the hearing aids, do they show?... If they are obviously immigrants or refugees then I will ask them about their country. (Participant 5)

I think doing a home visit is a really good way of [taking] a peek into the culture. I think home visits are generally a good idea, when you are first meeting a family. But I think particularly when you have a family from a different culture, you can learn a lot when you go to their home. So being respectful of their traditions, doing a home visit to find out more about their traditions. And also, I think home visit puts them at ease so they can talk a little bit more about themselves. (Participant 9)

Strategies during intervention. Strategies for different stages of the intervention process were proposed. These included explaining the health care system and modifying standardized language assessments and interventions. Describing the health care system involved not only explaining the services the practitioner provides, but also what is generally available to families from the health care system (e.g., coverage funding for hearing aids). Health care models vary across cultures, and immigrants may not be aware of what they can access.

It's really a learning experience to go and understand that here [in Canada] you can actually push and that you have rights to ask for more different things. So when the medical team is in front of you, they have no idea that you would not go and look for other options... So knowing that some people may not go look, it's important that they will be informed of every kind of choice they would have. And I think this is part of what is lacking in terms of being sensitive to the culture. Because here [in Canada], [if] they have little information, they [can] go look and they connect with other people. This is definitely not what would be done in some other cultures. If I look at an African family I have on my caseload - the kid is falling through the crack. The parents didn't ask [about what they have access to] and they were just following

the system and the kid had barely any support. And I think it is pretty typical of what could happen with [an] immigrant, they come and trust what is happening and then if the kid falls through the crack, they won't see it. (Participant 18)

When practitioners encounter challenges with standardized assessments, they use various strategies to overcome these challenges. This includes adapting standardized assessments by administering a subset from a battery of tests, informing caregivers and recording that the score does not present an accurate assessment of competencies, or using non-standardized checklists or vocabulary lists to gain insight into language abilities.

I will have them fill out like a 50-word beginning words that we have. And I'll have them fill out their understanding in their own language as well as English so that I have an idea of how the child is doing overall, you know, is the child learning a language - period? And then, you know kind of combining the two gives me an idea of how well the child is doing. (Participant 9)

Challenges in interventions were overcome by making an effort to use culturally sensitive materials and by tailoring the content to reflect family home environments. This included making decisions on the content of the therapy and even on the décor of the office.

You know I do a little garden with pudding and cookie crumbs and then those gummy worms when we're talking about gardens...this is a fun kind of activity of mine and again I would check with the families, tell them what I'd like to do, this has gelatine in it, are you okay with that, if they aren't, I'll find something else to do. (Participant 19)

I'm sensitive to using themes that are appropriate to the family. When I decorate the room I try to make sure that I...you know, I want to observe North American cultural preferences but I don't want to make them such that they are uncomfortable for something else.... Even asking parents, oh you know I understand Eid is coming, could you bring in some stuff and we can share it together. (Participant 13)

Characteristics of a Culturally Competent Practitioner

Practitioners described various characteristics of a culturally competent practitioner, with **Table 2** listing those described by all practitioners. The most common characteristics included being respectful of culture and language, having knowledge of cultural practices, and self-reflection.

Table 2**Characteristics of a Culturally Sensitive Practitioner**

Respects culture, languages

Knowledgeable of cultural practices

Reflects on own cultural identity, values, prejudices, biases, and assumptions

Open-minded

Modifies sessions (e.g., appointment times, content) when needed

Explains sessions to families in advance to determine if content is culturally appropriate

Engages in cross-cultural encounters (e.g., multicultural events)

Be respectful of culture and language. Many practitioners commented on the importance of being respectful of cultural practices. Respect involves acknowledging and accepting different cultural practices, thereby creating a safe space for families to receive services.

I think being respectful of cultural differences and value differences. I have a couple of adolescent Muslim girls who wear a hijab, respectful of practices, closing the door to the room when asking them to remove their hijab to work with the device. And I also realised that because that is their dress, garb, when I'm doing assessments in terms of hearing, I always do my assessments with them wearing their hijab because that's their typical wearing option. (Participant 7)

Well I think respect for the other cultures you're dealing with, as with any family, respect and trust have to be the basis of the relationship because we are asking them to buy in to the fact that their child with hearing aids or cochlear implants is going to learn to speak, and that is a leap of faith for most people, and if you've come from a culture or country where people who have hearing loss don't learn to speak, then it's even more of a leap. And so for them to believe you and to do what you are asking them to do at home, there has to be a foundation of trust in the therapist and so I try as hard as I can to create that trust and I think that begins with respecting their traditions and being open to whatever they want to tell me. (Participant 9)

Being respectful of the home language was also considered vital to culturally sensitive care. Practitioners noted on several occasions that they should be aware of a family's preferences to preserve their home language. Advocating for this approach was seen to help establish trust.

Also, respecting their language in terms of teaching it to the child. So most times, parents want their children to learn their own language. Because there's grandparents involved – because the parents themselves are very attached to their language. So they go ahead and do that and we do the therapy in English and they will translate to the child's language. I think it works well when there can be someone who speaks English and then another speaks the native home language so that the child really has a bilingual kind of setting. So to be respectful of their language. I mean, in our situation, because it's a language-based program – that's HUGE! (Participant 9)

So just to respect their culture and their language, because I don't want them to lose that as well, right? Cus I know that when I speak to parents they'll be like, oh we want them to...even if they don't understand fully the language they speak at home even if they are somewhat exposed to it. (Participant 17)

Have knowledge of cultural practices. Many practitioners also noted the importance of having some knowledge of their patient's cultural background prior to proceeding with service delivery in order to be sensitive to their needs. Strategies for learning about cultural practices include asking families about their day-to-day routines or describing the content of the therapy session in advance to see if everything is appropriate.

Culturally sensitive [care] would mean that you would want to have some knowledge of what the cultural practices are for that particular family. You would want to know their religious observances.... It would be being able to incorporate those things into the therapy, asking families you know "I'm doing it this way, how would you do that at home, what would be more appropriate as something I could help you with at home." (Participant 13)

I might also discuss with the families – well we do this anyways but – what are their routines at home, what types of toys they have and that's a big one because depending on the culture, they may or may not have the toys you expect them to have, so that the variety or the group of toys they may have could look very different... the types of songs they might sing or the types of play and routines they have could look very different too... so just not making assumptions around the fact that they are playing at home with the games and songs that I would expect to have in my house. (Participant 10)

Self-reflection. Nearly half of the practitioners commented on the importance of reflecting on personal cultural identity, values, prejudices, biases, and assumptions. Performing self-reflection was believed to help increase relationship-building opportunities and also establish trust.

If you're entering into the relationship without your own cultural biases...you're working with a family and you're accepting them...you're willing to listen to what their expectations are, what their needs are what they want to get out of the services that you're providing. What they see as their priorities. Not your priorities. It's really very key to that work. (Participant 16)

I always have to be mindful that my own culture...that's a bit different from the culture here...I have to be mindful that what I think could be pretty normal for me [but] may not be for everybody because we don't have exactly the same background.... (Participant 18)

Descriptions of other characteristics of culturally competent practitioners included being open-minded, flexible with appointment scheduling, modifying sessions when needed (e.g., appointment times, content), explaining sessions to families in advance to determine if content is culturally appropriate, and engaging in multicultural events (e.g., cultural festivities) to increase experiences with cross-cultural encounters.

Discussion

The aim of this study was to explore the experiences of practitioners with providing services to families of minority culture backgrounds. Practitioners encountered barriers throughout the process of service delivery with language barriers affecting every stage. Gender issues, lack of culturally sensitive materials, and Westernized language therapy programs occasionally presented challenges in service provision to families of minority culture backgrounds. Disabilities are also stigmatized in some cultures, which can present additional challenges to care

provision. Differences in child-rearing practices can also present challenges for family-centred services.

Research in the field of rehabilitation services has described barriers to providing care to families of minority culture backgrounds similar to what we found in the field of pediatric hearing loss. For example, language barriers have been noted to impact rehabilitation service delivery (Centeno, 2009; Dogan et al., 2009; Drolet et al., 2014; King et al., 2015) and male patients from some cultures sometimes explicitly request male practitioners (Al Busaidy & Borthwick, 2012; Dressler & Pils, 2009). Finally, cultural differences in language (verbal and nonverbal), play, independence, family structure, and perception of disability can also affect service provision (Al Busaidy & Borthwick, 2012; Cochrane, Brown, Siyambalapatiya, & Plant, 2016; Dressler & Pils, 2009; Lindsay et al., 2012; Pidgeon, 2015; Yang et al., 2006). Practitioners were able to mitigate these barriers with communication strategies, learning about cultural differences, explaining the health care system, and tailoring standardized assessments and therapy-based interventions when required.

Facilitators and strategies described by the participants in this study align with an international consensus paper on best practices in EHDI programs (Moeller, Carr, Seaver, Stredler-Brown, & Holzinger, 2013). This document states practitioners should be supportive of differing cultural beliefs and practices. Additionally, the document recommends practitioners to be reflective of personal discomforts and cultural biases as well as knowledgeable regarding how differing cultural practices can impact care.

Similarly, studies in rehabilitation have identified facilitators to culturally competent care that are consistent with our study. Practitioners seeking to learn about different cultural practices felt that asking patients about their values, beliefs, and daily routines helped to appropriately tailor care (Kinébanian & Stomph, 1992; Lindsay et al., 2012; Maul, 2010; Pidgeon, 2015; Stedman & Thomas, 2011). Although learning about cultures through education (e.g., attending lectures, workshops, or training) was noted to be a facilitator, it should be noted that this is not sufficient to becoming a culturally competent practitioner, especially with Canada's rapidly changing diversity. Training is limited as it cannot account for all cultures present in Canada. As a result, it is up to practitioners to go beyond what may be available at work, taking up more time and resources in an already busy schedule. Although we cannot offer a solution, future research on this important topic is warranted.

Existing literature supports modifying care to ensure cultural competence practices such as informal

assessments, translated materials, and considerations for material selection and culturally meaningful treatments (Cochrane et al., 2016; Pidgeon, 2015; Rhoades, 2014; Williams & McLeod, 2012).

This study also uncovered some key characteristics of a culturally competent practitioner. Key characteristics include practitioners being respectful, knowledgeable of cultures, reflective, open-minded, and tailoring care when needed. These characteristics align with the values of family-centred care, which advocates for partnerships between practitioners and diverse families (Rhoades, 2017).

Although culturally competent practices can help to improve patient experiences and outcomes, little information has been available about the barriers and facilitators that practitioners working in pediatric hearing loss have encountered in providing services to families of minority culture backgrounds. To the best of our knowledge, this is the first study to contribute to research on culturally competent care in EHDI services. Our study is not without limitations. It is possible that some examples described in this article may not accurately portray the values, intents, and practices of an entire culture. However, the goal of the paper was not to describe the culture but instead to portray how cultural differences can affect treatment. Other limitations include the study location; Canadian health care is publicly funded and therefore cannot represent global views. In addition, the setting of this study uses one particular service model, whereas other locations use different models. These limitations provide directions for future studies investigating cultural competence from the perspective of practitioners in pediatric hearing loss.

Increasing diversity can create challenges for EHDI services which are typically tailored to meet the needs of the majority population. Our study provides insight into barriers practitioners may encounter when providing services to families of minority culture backgrounds. Additionally, the findings in this study offer strategies that can be used to help overcome cultural challenges experienced by practitioners in hearing loss services. Future research in different provinces utilizing various service models across Canada may also help provide sufficient data to inform a knowledge-to-action intervention seeking to improve and maintain culturally competent practices in EHDI services.

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Appendix

Interview Protocol

Clinician Interview Protocol: Pediatric Hearing Loss Practitioners' Encounters with Cultural Minority Patient Families.

Purpose of Interview

I am meeting with practitioners who are or have provided services to minority culture caregivers of children with hearing loss in order to better understand service needs. I am defining *minority cultures* as any cultural groups that are not representative of the majority culture in Canada (e.g., caregivers that do not identify as primarily French or English Canadian). I am interested in learning about your experiences with servicing minority culture families. For example, I would like to hear about whether the delivery of care to this population is a smooth process or whether there are challenges. I would also like to hear about if/how you tailor care to suit the needs of minority culture families.

Procedure

I will ask you questions to guide our conversation but feel free to talk about your experiences and to add any information you feel is relevant and important. Please don't hesitate to ask questions. I'm going to start off by asking you some background questions, then I'll ask questions about your interpretation of culturally competent care. I'll then move on to asking you about your experiences servicing minority culture families and strategies that you might use to help improve service delivery.

Definitions

Cultural competence in a health care context has been defined as "understanding the importance of social and cultural influences on patients' health beliefs and behaviors; considering how these factors interact at multiple levels of the health care delivery system; and, finally, devising interventions that take these issues into account to assure quality health care delivery to diverse patient populations" (Betancourt, Green, Carrillo, & Ananeh-Firempong, 2003, p. 297).

General Information

Location of interview: Clinic

Other: _____

Gender: Male Female

Informant: Audiologist AVT

Other: _____

Education: _____

Years of practice: _____

Background Questions

What is your position title?

How long have you been working in this field?

Please tell me about your cultural heritage and the languages you speak.

Prompt: In the past year, what percent of your patients are cultural minorities? What are the most common cultural groups serviced? Again, cultural minority families are defined here as any cultural groups that are not representative of the majority culture in Canada.

What does the phrase 'culturally sensitive care' mean to you?

Prompt: In your opinion, what are key characteristics of a culturally sensitive practitioner?

Cultural Competence Questions

I'm looking for some information on the proportion of cultural minority families you service. In your current caseload, how often do you work with minority culture families? (e.g., most of the time, half of the time, some of the time, etc.)

Have you attended any courses or training on cultural competence?

Prompt: Did you receive any training in your degree? From your job?

Questions on Servicing Minority Culture Families: From Diagnosis to Intervention

Note: Some of these questions might be more relevant for a particular position in Audiology – if it's not part of your job, please describe any relevant encounter.

Did you encounter surprising reactions to the diagnosis? If you have, tell me about it.

Prompt: Did some cultural minority families perceive disability as something to be ashamed of or something to be concealed or as a gift?

Did you encounter challenges when discussing amplification options? If you have, tell me about it.

Prompt:

Did you have to use any strategies for hearing aid use? Did you feel that you had to do anything differently?

Were some opposed to amplification for cultural reasons?

Are there difficulties with achieving consistent amplification use?

Do some families seem to feel they need more guidance with amplification usage?

Did you experience challenges when providing therapy? If you have, tell me about it.

Prompt: Were some aspects of the therapy not applicable to the child's home environment?

Did you encounter challenges when filling out language assessment questionnaires? If you have, tell me about it.

Prompt: Were some aspects of the questionnaires not applicable to the child's home environment?

Personal Strategies for Working With Minority Culture Families

What are some of the methods you use when providing services to minority culture families?

Prompt: Do you modify standardized assessments such as administering a subset as opposed to a complete battery of assessments?

Prompt: Do you tailor therapy sessions in order to better reflect the patient's home environment?

Do you use any methods to help improve communication with minority culture families? If so, what are some examples? If not, please explain why.

Prompt: Did you try different strategies to improve communication like simplifying language?

What would you say is the most important factor in culturally appropriate care?

Prompt: Here are some examples: knowledge of cultural differences, knowledge of culturally sensitive practices, having culturally appropriate assessments and interventions, etc.

Is there anything you'd like to discuss that I haven't covered?



Perspectives of Parents of Minority Culture Backgrounds on Pediatric Hearing Loss Services: A Qualitative Inquiry



La perspective de parents issus de minorités culturelles sur les services offerts aux enfants ayant un trouble auditif : une recherche qualitative

KEYWORDS

CULTURAL COMPETENCE

PEDIATRIC HEARING LOSS SERVICES

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Abstract

Increasing diversity in Canada can create challenges for practitioners providing services to children with hearing loss. Culturally competent services are required to ensure appropriate care for our multicultural population; however, there is a scarcity of evidence in audiology to inform practice guidelines. The perspectives of families of minority culture backgrounds on the services their children receive could provide invaluable information for practitioners seeking to provide appropriate care for all their patients. The objective of this study was to explore minority culture families' perspectives on barriers and facilitators to culturally competent early hearing loss services. A qualitative research design with semi-structured interviews was employed. A total of 10 parents (representing nine different children) participated in this study. Three themes emerged from the interview data: experiences with hearing loss, services, and education systems; needs as a minority culture family; and helpful strategies for service provision to minority culture families. This study is one of the first to explore the experiences of minority culture families receiving early hearing loss services. Families who have children with permanent hearing loss often require long-term, ongoing, intervention services. For this reason, it is imperative for practitioners to provide culturally competent services informed by empirical evidence. Insights from this study offer a starting point for knowledge translation into clinical practice.

Editor-in-Chief:
David H. McFarland

Abrégé

L'accroissement de la diversité au Canada peut créer des défis pour les praticiens qui offrent des services aux enfants ayant un trouble auditif. Des services culturellement adaptés sont essentiels afin d'assurer des soins appropriés à une population multiculturelle. Cependant, peu de données probantes sont disponibles dans le domaine de l'audiologie pour orienter les guides de pratique. Les praticiens cherchant à fournir des soins appropriés à tous leurs patients pourraient tirer de précieuses informations sur les services offerts aux enfants issus de minorités culturelles à partir de la perspective des parents de ces enfants. L'objectif de la présente étude était d'explorer la perspective de familles issues de minorités culturelles quant aux obstacles et aux facteurs qui facilitent l'accès à des services précoces adaptés sur le plan culturel. Un devis de recherche qualitatif utilisant des entrevues semi-structurées a été sélectionné. Au total, 10 parents (représentant neuf enfants différents) ont participé à cette étude. Trois thèmes sont ressortis des données des entrevues : (1) l'expérience des parents avec le trouble auditif, les services reçus et le système d'éducation, (2) les besoins des familles issues de minorités culturelles et (3) les stratégies jugées utiles par les parents dans la prestation de services aux familles issues de minorités culturelles. La présente étude est l'une des premières à explorer l'expérience des familles issues de minorités culturelles sur les services précoces qu'ils ont reçus pour le trouble auditif de leur enfant. Considérant que des services d'intervention continus et s'étendant dans le temps sont souvent requis pour les familles qui ont des enfants ayant un trouble auditif permanent, il est essentiel que les praticiens fournissent des services adaptés sur le plan culturel qui sont guidés par des données empiriques. Les résultats de la présente étude offrent un point de départ pour le transfert de connaissances vers la pratique clinique.

Cultural competence in early hearing loss interventions is required to support language development amongst Canada's diverse population. The latest projections from the Canadian census indicate that by 2036, nearly half of the population will be immigrants or children of immigrants (Statistics Canada, 2017). In the United States, minority groups are anticipated to represent the majority of the population by 2044 (United States Census Bureau, 2015). As diversity increases in North America, early hearing loss practitioners face challenges related to the provision of services; specifically, services developed on Western norms limit the ability of practitioners to deliver culturally competent health services to a more diverse clientele (Bonilla & Edwards, 2011; Grandpierre et al., 2018, 2019). Culturally competent health care refers to

understanding the importance of social and cultural influences on patients' health beliefs and behaviors; considering how these factors interact at multiple levels of the health care delivery system; and, finally, devising interventions that take these issues into account to assure quality health care delivery to diverse patient populations (Betancourt, Green, Carrillo, & Ananeh-Firempong, 2003, p. 293).

There is extensive literature in rehabilitation to inform the development and maintenance of culturally competent services (see Al Busaidy & Borthwick, 2012; Jaggi & Bithell, 1995; Lee, Sullivan, & Lansbury, 2006; Lindsay, Tétrault, Desmaris, King, & Piérart, 2014; Nelson, Allison, & Copley, 2007; Williams & McLeod, 2012), yet a paucity of research in the field of audiology (Ball & Lewis, 2014; Eriks-Brophy, 2014; Grandpierre et al., 2018). Culturally competent health care services must be informed by evidence to ensure the best standards of care. As a family's cultural background can influence beliefs, values, behaviours, and practices, it is important for practitioners to be knowledgeable about cultural differences when providing services to families of minority culture backgrounds.

The diagnosis of a permanent hearing loss in a child is typically a stressful and emotional event for families without history of hearing loss. Yet, families of minority culture backgrounds are likely to encounter additional challenges beyond the hearing loss itself. For example, Canadian Indigenous children are more likely to experience poorer health determinants and living conditions than non-Indigenous children (Ball, 2008; Findlay & Janz, 2012a, 2012b; Statistics Canada, 2008). In addition, disability is stigmatized in some cultures and a diagnosis can bring shame upon family members, leading to relationships being severed (Jackson, Traub, & Turnbull, 2008). As a result, disabilities are often kept hidden.

Given the lack of research to inform best practices for delivering culturally competent care in pediatric hearing loss, the objective of this study was to explore minority culture families' perspectives on barriers and facilitators to culturally competent early hearing loss services. This study is part of a larger program of research seeking to gain insight into barriers and facilitators to culturally competent early hearing loss services from the perspectives of practitioners and minority culture families.

Method

Participants

Families of minority culture background receiving rehabilitation services for children with hearing loss were invited to participate from the Children's Hospital of Eastern Ontario (CHEO), a tertiary care hospital located in Ottawa, Canada, that provides diagnostic care as well as follow up and auditory-verbal intervention services. Clinicians who have previously worked at the Child Hearing Lab at the CHEO Research Institute also assisted with recruitment.

Caregivers were eligible to participate if they had (a) a child/children with permanent hearing loss who received early intervention services within the past 5 years and (b) a minority culture background, defined in this research as "any cultural groups that are not representative of the majority cultures in Canada." For families who did not speak English, a translator was provided. Eligible caregivers were approached by clinicians to inform them of the study. Clinicians determined eligibility with knowledge of the patients' backgrounds. Residency was also used to help identify potential participants as the Audiology Clinic at CHEO also provides services to Indigenous populations in Nunavut.

Abiding by qualitative research practices (Creswell, 2014; Kelly, 2010), a total of 6–10 participants was considered sufficient for gaining relevant insights into a subject that has received minimal attention. The Research Ethics Boards of the CHEO and the Université d'Ottawa approved this study (REB 16/01X).

Procedure

The method of *interpretive description* informed the data collection in this study (Thorne, 2016). Drawing on elements of grounded theory, naturalistic inquiry, and ethnography, interpretive description attempts to capture an account of a clinical phenomenon and make the results accessible to clinicians. Its foundation of qualitative inquiry is on a smaller scale in that it does not attempt to generate new theories, but instead produces a "tentative truth claim"

(Thorne, Kirkham, & O'Flynn-Magee, 2004, p. 6). That is, interpretive description provides a snapshot of a specific context at a specific time. This is typically achieved by drawing on data collected from small samples through methods such as interviews.

Data were collected via semi-structured individual interviews (see Appendix). Interview questions were informed by findings stemming from our scoping review on barriers and facilitators to cultural competence in rehabilitation services (Grandpierre et al., 2018). The interview consisted of basic sociodemographic questions (e.g., cultural and linguistic background, education, income, child's audiological information) as well as open-ended questions to help explore participant perspectives. Interviews were conducted by the lead author who has expertise in rehabilitation sciences and extensive experience in qualitative research. Medical records were used to supplement information on hearing loss (e.g., diagnosis, age of diagnosis, cause and degree of loss, amplification recommendations, and intervention information).

Field notes were taken by the lead author during and after interviews to record any overall impressions or general thoughts. Interviews were audio recorded and transcribed verbatim by two independent researchers. Field notes were consulted during analysis. Data were entered into NVivo (version 10.1.2), a qualitative software program used for coding. Demographic information was entered into SPSS (version 24), a statistics software program.

Analysis

Consistent with qualitative methods, analysis occurred concurrently with data collection (DiCicco-Bloom & Crabtree, 2006). Inductive data analysis techniques were used to identify themes and commonalities (Thorne, 2016). The lead author analyzed the transcripts using constant comparative method, a coding process based on Corbin and Strauss's (1998) open, axial, and selective coding methods that is consistent with interpretive description methodology (Thorne et al., 2004). During open coding, each passage in the transcripts was assigned a label. In axial coding, characteristics of each label were then compared between interviews to facilitate consistency and eliminate redundancy. Finally, similar labels were categorized and then collapsed into major themes during selective coding.

Trustworthiness (Krefting, 1991) was established to ensure transparency and quality. Trustworthiness consists of four components: credibility, transferability, dependability, and confirmability. Credibility was

achieved by consulting coauthors regarding analysis and interpretation. The provision of in-depth details in the data, along with numerous supporting quotes, ensures transferability. Clear and detailed steps documenting the research process help to ensure dependability. Finally, recording and consulting field notes as well as consulting coauthors with expertise in pediatric hearing loss, health sciences, medical research, and qualitative research help to ensure confirmability.

Results

Context

The study took place in a setting where newborn hearing screening has been implemented province wide. In Ontario, Canada, infants receive screening for hearing loss after birth. Children with results that indicate the need for referral are then sent for diagnostic audiological assessments. Following a diagnosis, audiologists will discuss hearing technology options with families. Discussions of language intervention approaches (e.g., spoken language approaches, visual approaches, and total communication approaches) and financial resources are typically provided by a family support worker. The intensity of language interventions can vary. Children are typically seen weekly for listening and spoken language intervention during the preschool years. During the school years, they are generally seen once to several times weekly based on their needs as determined by the specialist in language intervention.

Description of Participants

A total of 10 parents (representing nine different children) were interviewed. The mother and father of one child chose to be interviewed separately. Half of the interviews were conducted on the phone as per parent requests with the remainder conducted in person. The average interview length was 27.55 minutes (range = 17.01–47.47 minutes).

Table 1 provides a description of child characteristics. Three children did not receive screening due to the lack of a screening program in their country of origin. Median age of hearing loss confirmation was 5.1 months (interquartile range = 4.8–12.5). All children had bilateral hearing loss, were aided with hearing technology (e.g., hearing aids, cochlear implants), and received auditory-verbal therapy. Etiology was known for six of the nine children. All children were receiving or had received auditory verbal therapy. The children had a mean age of 4.8 years (standard deviation = 2.6) at the time of the interviews, and, as such, parents had over three years of experience with hearing loss.

Table 2 provides characteristics of the caregiver participants. Three families had immigrated to Canada within the past 3 years; two had lived in Canada between 10–17 years, and the remaining, over 20 years. Most of the caregivers had university and post-graduate degrees ($n = 7$). Just over half ($n = 5$) reported an annual family income of \$60,000 or over; for context, the median household income in Canada is approximately just above \$80,000 (Statistics Canada, 2019). One family chose not to disclose their income.

Thematic Results

The goal of the interview was to gain some insight into minority culture caregivers' experience receiving early hearing loss services in Ontario, Canada. Three themes emerged from the interview data: experiences with hearing loss, services, and education systems; needs as a minority culture family; and helpful strategies for service provision to minority culture families.

Table 1	
Child Characteristics	
Characteristics	Children ($n = 9$)
Sex, n (%)	
Female	3 (33.3%)
Route to confirmation of hearing loss, n (%)	
Screened	6 (66.6%)
Passed	1 (11.1%)
Referred	5 (55.5%)
Age at confirmation, (months), median (IQR)	5.1 (4.8, 12.5)
Onset of hearing loss, n (%)	
Congenital	3 (33.3%)
Early onset	2 (22.2%)
Late onset	1 (11.1%)
Unknown	3 (33.3%)
Hearing loss description, n (%)	
Bilateral	9 (100%)
Degree of hearing loss, n (%)	
Moderate	3 (33.3%)
Mod-severe	2 (22.2%)
Severe	1 (11.1%)
Profound	3 (33.3%)
Amplification, n (%)	
Hearing aids	3 (33.3%)
Cochlear implants	6 (66.6%)
Etiology, n (%)	
Known	
NICU graduate*	2 (22.2%)
Hereditary/genetic	2 (22.2%)
ENT malformation	2 (22.2%)
Unknown	3 (33.3%)
Age at interview (years), mean (SD)	4.8 (2.6)

Note. *NICU does not include children with syndromic hearing loss or ENT anomaly.

ENT = Ear, nose, and throat; IQR = interquartile range; NICU = Neonatal Intensive Care Unit; SD = Standard deviation.

Table 2	
Parent Characteristics	
Characteristics	Participants (n = 10)
Interviewee, n (%)	
Mother only	9 (90%)
Father only	1 (10%)
Languages spoken at home, n (%)	
English	9 (90%)
French	3 (30%)
Chinese	1 (10%)
Arabic	4 (40%)
Somali	1 (10%)
Russian	2 (20%)
Creole	1 (10%)
Countries of origin, n (%)	
Lebanon	2 (20%)
France	1 (10%)
Philippines	1 (10%)
Cambodia	1 (10%)
Somalia	1 (10%)
Syria	1 (10%)
Haiti	1 (10%)
Iraq	1 (10%)
Africa	1 (10%)
Ukraine	2 (20%)
Highest level of education in household, n (%)	
None	1 (10%)
College/university	9 (90%)
Family income, n (%)	
Below \$20,000	2 (20%)
\$20,000 to less than \$40,000	2 (20%)
\$60,000 to less than \$80,000	2 (20%)
More than \$80,000	3 (30%)
Did not disclose	1 (10%)

Experiences with hearing loss, services, and education systems.

Perception of hearing loss. When asked about how their cultures perceive hearing loss and disability, many of the parents described how it is stigmatized, taboo, and often hidden. For some, this stigmatization resulted in them not disclosing the hearing loss to close friends or family.

Disability back at home...it's like you put [a disabled person] into an institution and lock the door. No

one sees them, no one hears about them, nothing... When (child's name) first started to wear her hearing aids, we couldn't take her [out]...like when we went to certain places, we couldn't put the hearing aids on. Our Lebanese culture...they don't understand the fact that you know, I need glasses to see. It is like "oh my god, she is disabled!" So, there was a little bit of a struggle with some of our family members - but at the end, (husband's name) and I, were like whatever, this is our kid. If she needs it [hearing aids], she is going to wear it.

We don't care if they don't like it, you don't have to see us. (Participant 1)

We don't share a lot. We are very private. I mean private - like not even my friends are supposed to know [about disabilities or disease]. So, it is something you are not able to control...hearing loss, autism, all of this is taboo. (Participant 7)

When exploring reasons for why hearing loss is stigmatized, some parents described a lack of knowledge surrounding hearing loss and the need for hearing technology interventions.

There is no one with hearing loss [where participant grew up].... Ones that do have hearing loss, it's not like in Canada, they are always put in to this separate little category.... So, when we say he has hearing loss, they [family] are like oh are you sure? Have you tried this and that? Like it is hearing loss, he can't hear us properly! (Participant 3)

Because there are not a lot of children hearing aids in our home country.... They wanted to put all the children in the special schools, so in real life there is no children with hearing aids here [Ukraine] at all. (Participant 10)

All parents reported experiencing a wide range of emotions when learning of their child's diagnosis, from relief to having the loss finally confirmed, to shock, devastation, denial, and even seeing it as a divine gift.

My reaction was the same as my family's in the beginning, I was just shocked. But at the end of the day, we're people that are spiritual - so we do understand that God creates people differently. And it's a gift that was given to her, only to her, and we accept it. (Participant 5)

Perception of services. Following the diagnosis, almost all parents (90%) reported they felt happy with the services their children received. The majority felt that practitioners were sensitive to their cultural needs. Practitioners were able to fully address their concerns and gaps in knowledge regarding hearing loss and technology, while also ensuring they understood intervention options.

You have no idea how lucky I feel to have that kind of team.... Like the team were absolutely amazing. Like they gave us everything, they explained everything. We came out of our few meetings, very aware of the situation and they were always there to help us out if we had any questions.... Honestly, I would not have been able to keep going without our team. I would tell our speech therapist, "I can't have [child's name] wear the hearing aids all time

because of the people [from her country]" and then she also gave me some therapy, she made me feel so much better coming out of it. (Participant 1)

Another parent noted, "They are extremely happy with the services at CHEO because it is better than they ever expected" (Translator for Participant 8).

However, a few families emphasized their wish to raise their child multilingually and felt that those needs were not met. They stated how it was a struggle to decide on which language to focus on (e.g., service language vs. home language). Some also expressed the desire for more multilingual practitioners.

The first thing the audiologist told us was to concentrate on one language and one language only because he had hearing loss. To us, that is important to our family because we speak Arabic at home.... Because had we not stuck to it, and he would only have spoke English and he wouldn't have a way to communicate with his grandparents or great-grandparents. (Participant 3)

It was this whole question of how to integrate the whole family...so I don't see [how] you can pick one [language] or the other. And so, they actually got us to meet a bilingual therapist.... She was really realistic that it may be a challenge for a child with hearing loss, but she was able to help us to actually try to teach him two languages. And I think this is because we are in Ottawa. I just talked recently with someone saying that the family who has a little boy who is only 5, they were told that they should pick one language and they should actually pick English. (Participant 2)

Perception of education systems. Finally, several parents expressed an appreciation for the option to include their children in the general education system. They explained that, in their home country, children with disabilities reside in institutions. Parents even experienced challenges when registering their children into weekend schools where their home language is taught: "When a child has something that is out of the norm, they are usually institutionalized. They are usually put in to a special center. So a child like mine would definitely be in a special school" (Participant 2).

In Ukraine, it was horrible, yes. Every half a year, we had the medical commission [medical examination]. Every time they told they need to put him to special school [institution]. So every time I refused.... Here [Canada], nobody told me to send my child to a prison for children (laughs, referring to special school). Everyone just wants

to help him, speak and understand. And everyone wants to help me as well and it is really great!! (Participant 3)

I was looking to register them [her children] for Arabic school on Saturday, because they can speak fluently but I want them to read and write it and I filled out the online registration form and then the person from registration called me...cultural differences sometimes, they are brutal. [Child's name] has two cochlear implants. I said the teacher would have to wear a mini-mic so her voice would go directly to his processors so he is able to hear her clearly. And she goes, "oh! Well I have never had such a...something as hard and different as your son's case!" And I am like, hard and different? Are you saying strange? You speak as you haven't come across it.... It is not as hard as you make it sound. He is fully functional, learning three languages! I think that's when cultural differences lead to restrictions. (Participant 3)

Needs as a minority culture family. The majority of the parents described how vital it was to have access to a translator when language barriers were present. Parents who recently immigrated to Canada, did not know the service languages, and had a child that required ongoing medical attention emphasized the need to have the same translator at each appointment.

They [the parents] can't understand anything that the doctors are saying. So to them, the interpreter is the doctor. From their experience, it's very frustrating [that] they can't understand the medical staff. Extremely frustrating. It should be the same interpreter that follows with the family. Because he says every time they have to bring a different interpreter, they have to say his story all over again. He also said that he wants to have access to the interpreter. He wants access so he can call the interpreter and interpreter can call the hospital and speak to them. (Translator for Participant 8)

Explanations of the health care system were also seen as crucial to culturally competent service delivery as many parents emphasized how different health care is in their home country. Some deeply appreciated explanations from practitioners about what is and is not covered by Canadian health insurance, as well as what supports and services are available. Others felt such explanations need to be routine.

In Ukraine we get nothing from government.... It is absolutely different experience. We bought everything by ourselves, with the help of my family, with the help of my parents.... We had to buy the hearing aids and everything by ourselves. Here, when we arrived here, they [practitioners] explained everything, there are lots

of programs that can help the families with children with the disabilities, there is a social worker. Everybody was so kind, so polite, so friendly, it is great. Really great! It is really wonderful, especially for children and especially for children like [child's name]. (Participant 9)

...what I found was the kid is falling through the cracks. The parents [friends of participant] didn't ask [about support availability] and they were just following the system and the kid had barely any support. And I think it is pretty typical of what could happen with immigrant...they come and trust what is happening...and then if the kid falls through the crack, they won't see it. (Participant 2)

Parents also expressed a desire for practitioners with experience with multicultural families. They felt that practitioners with more experience were better able to provide care that was culturally sensitive and could be tailored to meet the needs and values of their family.

But I think again it goes back to the professional themselves, if they have had the chance to work with different clientele and different cultures, then they are more open to other peoples' ideas. For others who generally dealt with certain clientele - they don't tend to be as open-minded. Some people are not open-minded because it's just black and white. For some, they are set in their ways and they are not as willing to listen to others and see things for what they truly are. (Participant 3)

Another participant noted, "Being sensitive to somebody's needs [when asked what's important for culturally sensitive care]. Just understanding [cultural differences] I guess. Patience and understanding is pretty much all it is" (Participant 6).

Helpful strategies for service provision to minority culture families. Parents described a variety of strategies practitioners used to help them. This included providing reading materials, ongoing support, and visual cues when language barriers arose. For one of the families, the parents did not believe that their child had hearing loss until the practitioner conducted a hearing test and gave them the same test.

He [father] was convinced the second time when he [in a hearing test] actually heard the sound in his own ears and he couldn't take it because it was so loud. Whereas for his son, he wasn't even annoyed by it. He was just playing. So, the father was convinced. (Translator for Participant 8)

Parents also reported how helpful it was to have practitioners that patiently provided clear and thorough

explanations of the hearing loss and what was to be involved in the intervention. Another strategy was repetition, as parents are not just struggling with language barriers but also the emotional trauma of learning how to navigate life with a child with a disability.

They told me about the ABR [auditory brainstem response] tests, and the audiogram and how to read it. That was very helpful. Then they told me that she needs hearing aids and they taught me how to put on and take off these hearing aids and how they work. They taught me how.... I was taught to basically train her to talk and what - to this day, we attend speech therapy tries to help us to...focus more on her and try to teach her how to talk, and different activities and ways to communicate with her and try to encourage her to talk. I found all that helpful. (Participant 5)

Example Case Study

This section provides a case study of one interviewee, with all identifying information changed to ensure confidentiality. This case helps capture a detailed account of experiences families of minority culture background may encounter.

Akram and Amira are Middle Eastern refugees who immigrated to Canada in late 2016 with their daughter, Maya. Maya is two years old and only had her hearing loss discovered upon her first health checkup in Canada. There is no newborn hearing screening in her home country. Her parents had stated they did not believe she had hearing loss for a long time. It was only when their daughter's audiologist showed them pictures of the cochlea, explained the type of loss she had, and pointed out how their daughter's younger brother had more words than her that they began to believe the diagnosis.

With the help of a translator, the audiologist discussed the ways in which their daughter would benefit from bilateral cochlear implant surgery. She also discussed other intervention options, such as sign language, to ensure that they could choose an intervention that aligned with their values. Akram and Amira initially felt afraid of the surgery. They communicated this fear and, in response, the audiologist showed them pictures of the ear and explained the surgical process.

Though access to a translator helped overcome their language barriers, Akram stated that the pictures helped them better understand what was involved in the surgery as they were not familiar with medical terminology in their own language. They expressed extreme gratitude

for this strategy as it enabled them to make an informed decision. Maya now receives weekly auditory verbal therapy, a spoken language approach to language therapy. Her language therapist speaks fluent Arabic and, as a result, she tailors all the therapy to their language and culture, thereby ensuring culturally sensitive services. Akram states that, in this way, everything (e.g., therapy content) is relevant and helpful.

Discussion

The aim of this study was to gain insight into barriers and facilitators to culturally competent early hearing loss services. Specifically, the goal was to explore minority culture caregivers' experiences with services for their children.

A child receiving a permanent hearing loss diagnosis was the beginning of many cultural challenges that families encountered. Discussing the diagnosis and amplification needs with family members and friends was often described as difficult due to cultural stigma. An examination of existing literature suggests perceptions of disability vary across cultures and can impact the way families experience and manage the diagnosis (Jackson et al., 2008; Zhang & Bennett, 2003). Though these views did not influence every parent's experience, it is important for practitioners to be aware of these different perceptions when providing services to newcomers. Minority culture families may have specific cultural and linguistic needs and values that practitioners need to be aware of and address to ensure culturally competent care delivery.

Language barriers also created stressful experiences for families unable to converse in English or French. Access to a translator was seen as beneficial, but having access to the same translator who is familiar with the child's medical history can relieve additional stressors for the parents. Other helpful strategies for facilitating communication include providing thorough explanations with repetition to ensure comprehension. This is consistent with Yeowell's (2010) study in which a participant of minority culture background recommended practitioners talk slowly and conduct comprehension checks by asking the patient to repeat back the information told to them.

Families in the current study also reported experiencing challenges due to practitioner perceptions of multilingualism. Specifically, they voiced a need for practitioners to promote multilingualism. Although many challenges were reported, several families felt appreciative of the services they received and also provided recommendations for improving culturally competent

services. Such recommendations included providing clear explanations of the diagnosis and interventions, and to supplement explanations with additional reading materials and ongoing support. Descriptions of the healthcare system (e.g., coverage, support services) were also seen to be crucial for families who had recently moved to Canada to address the sometimes strong contrasts between what is available in Canada vs. their home countries. The need for explanations of health care systems has also been documented in several other studies, highlighting the need for a change in health care practices when servicing minority culture families (Kummerer & Lopez-Reyna, 2006; Nelson et al., 2007; Yeowell, 2010).

Practitioners experienced in delivering services to multicultural patients were also viewed as valuable assets to culturally competent care. When this is not possible, seeking knowledge on a patient's cultural background, attending workshops on cultural competence, or even attending cultural events can be beneficial to practitioners (Lindsay et al., 2014; Nelson et al., 2007; Stedman & Thomas, 2011). Our recent systematic review (Grandpierre et al., 2018) included additional recommendations, such as communication strategies (using simple language, speaking slowly, using visual aids such as pictures, etc.).

More research is needed to inform culturally competent practices in early hearing loss services. A limitation to our study is that our participants were selected from one setting with a particular service model. In addition, only three of the families were recent newcomers, thereby further limiting insights into experiences of minority culture families. There is also a lack of insight from Canadian Indigenous families and less educated families. However, the limitations in this study provide direction for future research investigating the perspectives of families of minority culture backgrounds receiving hearing loss services for their children.

This study is one of the first to explore the experiences of minority culture families receiving early hearing loss services. Families who have children with permanent hearing loss often require long-term, ongoing, intervention services. For this reason, it is imperative for practitioners to provide culturally competent services informed by empirical evidence. Insights from this study offer a starting point for knowledge translation into clinical practice.

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Appendix

Interview Protocol

Parent Interview Protocol: Minority culture parents of children with permanent hearing loss

Purpose of Interview

I am meeting with minority culture parents to help me understand what your service needs are.

I would like to hear about your experiences with your child’s hearing loss services. Specifically, I would like to hear about how you learned about your child’s hearing loss, what your experience was like when you were told about the treatments available, and what your experience was like with the language therapy sessions.

Procedure

Before we begin, I’ll ask you some general questions about your child’s hearing loss. Next, I will begin asking you questions to guide our conversation, but feel free to talk about your experiences and to add any information you feel is important. Please don’t hesitate to ask questions. I’m going to start off by asking you some background questions. Next, I’ll ask about how you found out about your child’s hearing loss, and then I’ll ask about your experiences with receiving hearing loss services.

General Information for Interviewer

Location of interview: Home Clinic Other: _____

Informant: Mother Father Other: _____

City of residence: _____

Number of children: _____ Number of children with hearing loss: _____

Age of child/children: _____ Age of diagnosis: _____

Amplification: Hearing aids Left ear Right ear

Hearing aids Left ear Right ear

Age of amplification: _____

Background Questions

Have you always lived in Canada?

Tell me a little bit about your family.

Prompt: Are your kids in school yet?

Tell me about your family’s cultural background.

Prompt:

What is your child’s/children’s cultural heritage?

Do you have any cultural traditions?

Tell me about something about the customs of your culture (e.g., meals).

Are there any differences in the health care system?

What languages do your family speak?

Hearing Loss History

Tell me about how you found out about your child’s hearing loss.

Prompt:

Was your child screened at birth?

Tell me about the process from screening to when your child’s hearing loss was confirmed. How many visits?

How long was it before you got the confirmation that your child had a hearing loss?

What were your needs from the time that you learned your child potentially had a hearing loss to after the diagnosis?

Prompt:

What kind of information did you find helpful in the beginning?

What information or guidance did you need after the diagnosis (e.g., after confirmation)?

What kind of supports did you need, e.g., social worker, therapist, family?

What supports/information did you receive?

Cultural Information

How is hearing loss regarded in your family's culture?

Prompt:

Is hearing loss viewed as a disability? Is it viewed negatively or positively?

How are disabilities viewed/seen in your culture?

What was your reaction to discussing and receiving hearing aids, assistive hearing devices, or cochlear implants for your child?

Prompt:

Did you initially want your child to have hearing aids/assistive technology/cochlear implants? Why/Why not?

Sometimes parents have a hard time with having their child's hearing loss being made visible. Tell me about your experience.

Experiences with Hearing Loss Services

Now I'm going to ask you about what you thought of your child's hearing loss services from diagnosis to the time you received treatments. Would you have wanted information on how Canadian health care works?

How did you feel about using English/French in your hearing loss appointments?

Prompt:

Would you have wanted an interpreter?

Were you encouraged to use your language with your child? How did you feel about that?

How did you feel about how your health care practitioner told you about your child's hearing loss?

Prompt:

Was your doctor/therapist sensitive to your experience?

What would be the best way to be told about your child's hearing loss?

How did you feel about the way your health care practitioner discussed hearing aids, cochlear implants, or assistive hearing technologies with you?

Prompt:

Was your health care practitioner sensitive to your needs?

Was your health care practitioner sensitive to your values?

If you could improve this experience, what would be different?

How do/did you feel about your child's language therapy?

Prompt:

Were the materials used in your child's language therapy items that you are familiar with?

What did you think of the therapy that you needed to continue at home?

What are some recommendations for improvement?

When you've filled out questionnaires that look at your child's hearing and language skills, did you find the content describing your home environment?

Prompt: Did the questions and answers work with you and your child's experiences or did you find some things that didn't work? For example, were some parts discussing things that don't exist in your home environment?

What would you say is the most important thing for good patient care when practitioners and patients don't share the same language?

Is there anything you'd like to discuss that I haven't covered?



Good Vibrations: A Proof-of-Concept Study of the Preferred Temporal Characteristics in Surf-Like Sounds for Tinnitus Therapy



Les bonnes vibrations : une étude de démonstration de faisabilité des caractéristiques temporelles des sons ressemblant à ceux du surf à privilégier pour le traitement des acouphènes

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Abstract

A common audiological method of tinnitus management is to reduce tinnitus audibility by masking. To be effective, masking sounds need to be comfortable for long periods of time. Nature sounds, such as ocean waves or surf sounds, have been suggested to be effective for this reason. Natural or simulated surf sounds are typified by rhythmic oscillations in intensity. There are established asymmetric behavioural and physiological responses to oscillatory sounds that are ramped (gradually increase in intensity then decrease rapidly) versus damped (increase rapidly then decrease gradually over time). Ramped sounds engage attention while damped sounds are more comfortable. The aim of this study was to determine if such asymmetries in response are also translated to tinnitus masking. Two experiments were undertaken with groups of 10 tinnitus sufferers. In Part 1, an experimental round-robin tournament method was used along with rating scales to compare preferences among four recordings of natural surf sounds. In Part 2, a round-robin tournament comparing nine simulated surf sounds was used. Results indicated a preference for damped sounds over ramped sounds. Slower oscillations (rise and decay times of 5–8 seconds) were preferred to faster oscillations (rise and decay times of 2 seconds). The asymmetry in short-term tinnitus masking response to ramped and damped sounds is consistent with existing psychoacoustic research. The potential clinical use of oscillatory sounds and mechanisms underpinning observations are discussed.

Abrégé

Une approche fréquemment utilisée en audiologie pour le traitement des acouphènes est le masquage par un son, ce qui réduit la sensation de l'acouphène. Pour être efficaces, les sons masquants doivent pouvoir être écoutés confortablement sur de longues périodes. C'est pour cette raison que les sons de la nature, comme les sons de vague ou de surf, ont été suggérés comme étant efficaces. Les sons de surf, naturels ou simulés, sont caractérisés par des oscillations rythmiques variant en intensité. Il y a des réponses comportementales et physiologiques asymétriques connues aux sons dont l'intensité augmente graduellement puis diminue rapidement et ceux dont l'intensité augmente rapidement puis diminue graduellement dans le temps. Les premiers attirent l'attention tandis que les deuxièmes sont plus confortables à écouter. L'objectif de cette étude était de déterminer si ces réponses asymétriques se traduisaient également par une diminution de la sensation des acouphènes. Deux expériences ont été réalisées auprès de groupes composés de 10 personnes ayant des acouphènes. Dans la première expérience (*Part 1*), une méthode expérimentale d'essais circulaires et des échelles de cotation ont été utilisées pour comparer les préférences des participants entre quatre enregistrements de sons de surf naturels. Dans la deuxième expérience (*Part 2*), un essai circulaire comparant neuf sons de surf simulés a été utilisé. Les résultats indiquent que les participants ont une préférence pour les sons dont l'intensité augmente rapidement puis diminue graduellement dans le temps. Les résultats indiquent également que les participants préfèrent les oscillations plus lentes (temps de montée et de descente entre 5 et 8 secondes) aux oscillations plus rapides (temps de montée et de descente de 2 secondes). Les réponses asymétriques observées lors de courts masquages des acouphènes par des sons dont l'intensité augmente graduellement puis diminue rapidement et des sons dont l'intensité augmente rapidement puis diminue graduellement dans le temps sont consistantes avec les résultats d'autres recherches en psychoacoustique. L'utilisation clinique potentielle des sons oscillatoires et les mécanismes qui sous-tendent les observations ayant été effectuées sont discutés.

Tinnitus is a common audiological complaint that is often managed by a combination of instruction, counselling, and sound therapy (Hoare, Searchfield, El Refaie, & Henry, 2014; Searchfield, Durai, & Linford, 2017). Tinnitus “sound therapy” is a catch-all name for therapies using sound to reduce tinnitus. Sound therapies include total and partial masking (Tyler, Noble, Coelho, & Ji, 2012), relaxation (Davis, Paki, & Hanley, 2007), desynchronization (Eggermont & Tass, 2015), tonotopic reorganisation (De Ridder, Vanneste, Engineer, & Kilgard, 2014), and other putative mechanisms (Searchfield et al., 2017). Although masking has its roots in observations of the effects of natural sounds on tinnitus (Stephens, 2000), its recent history began with the psychoacoustical observations of Feldmann (1971, 1981) and the development of wearable maskers by Vernon (1981). Forty years after its emergence as a clinical method for managing tinnitus, there is still debate as to the most appropriate level (Jastreboff, 1999; Tyler et al., 2012) and type of sound to treat tinnitus (Hoare et al., 2014; Searchfield et al., 2017), and mechanisms of tinnitus masking are still uncertain (Hoare, Adjamian, Sereda, & Hall, 2013).

Masking sound preferences are likely to vary from person to person and may be due to personality, memories, and context (Searchfield, 2014). Traditional masking sounds have tended to be steady state broadband noise. Broadband noise tends to be well tolerated, void of meaning, and stimulates a wide bandwidth of hearing—all factors thought to assist in habituation to sound (Jastreboff, 1999). Tone-based therapies have been developed as attempts to desynchronize or remap frequencies putatively involved in tinnitus perception (Eggermont & Tass, 2015). Natural environmental sounds, and synthesized copies, have recently re-emerged as sound therapy as digital signal processing, digital memory in ear level devices, and wireless streaming have become widespread (Barozzi et al., 2016). Although clinical trials do not demonstrate superiority of natural sounds over broadband noise in the medium term (Barozzi et al., 2016), there is some evidence of patient preference for temporal variation in sounds in the short term (e.g., Henry, Rheinsburg, & Zaugg, 2004), and it has been suggested that the mode of effect for natural sounds may be different from broadband noise over time (Durai & Searchfield, 2017). The effect of noise may be primarily due to a “presence of sound effect” masking tinnitus, while the effects of natural sounds may be a result of their influence on emotion (Durai & Searchfield, 2017).

Temporally varying noise with oscillations resembling ocean waves or surf sounds have become common options within hearing aid manufacturers’ tinnitus treatment

devices (Sereda, Davies, & Hall, 2017). The sounds may aid relaxation; however, the benefit and mechanism of effect is unclear. The rationale underpinning the selection of waveform and oscillation characteristics is very subjective. The importance of oscillation characteristics on normal audition is far from trivial (Bach, Neuhoff, Perrig, & Seifritz, 2009; Tajadura-Jiménez, Väljamäe, Asutay, & Västfjäll, 2010), yet little effort appears to have been made to investigate established psychoacoustical and emotional consequences of varying sound oscillation patterns on tinnitus (Reavis et al., 2012).

In the context of this study, oscillating sound refers to periodic changes in amplitude of the temporal envelope of natural or synthesized broadband noise. We deliberately exclude modulations in tones that are unlikely to be effective tinnitus maskers, but may disrupt tinnitus through other mechanisms (Reavis et al., 2012). We focus on sounds that oscillate in such a way as to be perceived to sound like ocean wave “surf” sounds. These sounds contain a wide frequency range and oscillate over time with an initial increase in intensity, a steady state (plateau) at maximum intensity, and then a decrease in intensity. A noise that oscillates over time with a rapid increase in intensity followed by a longer duration decrease in intensity over time is defined as *damped*. When a noise increases in intensity incrementally, but decreases are large and abrupt it is defined as *ramped*. Sounds that have equal rise and fall are defined here as *symmetrical*.

Classical examples of the use of variation in oscillation characteristics exist in music (Huron, 1992). In music, increases of stimulus intensity level are more effective than equivalent decreases in gaining listeners’ attention (Huron, 1992). It is not possible to do this across an entire piece of music so instead composers have applied ramps where intensity increases are gradual, but stimulus decreases are large and abrupt (Huron, 1992). The temporal envelope of many sounds in nature are asymmetrical (Schlauch, Ries, & DiGiovanni, 2001). There may be an evolutionary reason for our response to ramped sounds, a perceptual bias to looming sounds may have provided a natural selection advantage as these sounds are perceived as approaching the listener and therefore have greater salience (Bach et al., 2009; Neuhoff, 2001; Tajadura-Jiménez et al., 2010). Ramped sounds have been argued to have greater saliency and elicit responses that would be expected to sounds containing warning information (Bach et al., 2009; Tajadura-Jiménez et al., 2010). Attention, and then response, to ramped sounds may have been an important factor in our survival. Such reactions are still observed in our modern environment; for example, we react when we hear a siren

signalling the approach of an emergency vehicle. This perceptual bias for ramped sounds is further illustrated by the finding that there is overestimation of the change in loudness of ramped sounds, and is thought to be an important adaptive mechanism, since underestimating the actual distance of a sound source could provide the listener with a selective advantage (Neuhoff, 2001).

Ramped sounds have been shown to elicit greater skin conductance as a measure of alertness, and their perceived loudness is greater and they sound longer in duration (Bach et al., 2009). Ramped sounds have been rated as more unpleasant and arousing than damped sounds that appear to recede from the listener (Bach et al., 2009). This effect appears most clearly expressed in response to unpleasant sounds, but may not exist for pleasant or neutral sounds (Tajadura-Jiménez et al., 2010).

The purpose of this study was to evaluate patient preference for various recorded natural surf sounds and to create and compare simpler synthesized versions with varying temporal characteristics (i.e., different rise and fall time, plateaus, and speed of oscillation). To improve sound therapy effectiveness, we need to explore the various parameters of sounds used and factors leading to success or failure amongst individuals. The terms *proof-of-concept*, *pilot*, and *feasibility* are sometimes used interchangeably when referring to types of studies, but they are different. The use of these terms has been criticized in some publications as a mechanism to explain poor study design, or in particular, small sample size (Arain, Campbell, Cooper, & Lancaster, 2010). The different study designs have important roles in research (Eldridge et al., 2016). There are slightly divergent definitions of what a proof-of-concept study is depending on the field of research. In medical research, feasibility and proof-of-concept are often used interchangeably. In technology development, proof-of-concept is used as a mechanism alongside rapid prototyping to quickly ascertain the relative merits of concepts before development into a form that can be applied in feasibility or pilot studies (Kendig, 2016).

In this study, an engineering definition of proof-of-concept was used: initial data from a small number of tests to validate and inform the continual development of a technology (EPSRC, 2015). Our purpose was to evaluate possible sound therapy stimuli to focus efforts on the most promising temporal characteristic(s). It was hypothesized that there would be a preference between sounds for tinnitus management based on their temporal characteristics. It was proposed that damped sounds would be preferred from ramped sounds as tinnitus maskers.

Method

The University of Auckland Human Participants Ethics Committee approved the methods used in this study (protocol 7928). Participants were recruited from a tinnitus research volunteer database and were required to have continuously present tinnitus. All participants received an information sheet briefly describing the study and provided written informed consent. The research was undertaken in two parts. Part 1 was an experimental round-robin tournament along with rating scales to compare listeners' preference among four recordings of natural surf sounds. Part 2 was the synthesis and round-robin comparison of simulated surf-like sounds that had varying rise and fall times.

Procedures

Prior to the experiments, to enable their tinnitus experience and demographics to be recorded, participants were asked to complete a Tinnitus Case History Questionnaire (Langguth et al., 2007) and the Tinnitus Functional Index (Meikle et al., 2012).

Otoscopy (Welch Allyn 3.5 V Diagnostic Otoscope) was followed by pure-tone audiometry undertaken using the modified Hughson Westlake procedure (Carhart & Jerger, 1959) in a sound booth (ISO 8253-1) with an Otometrics Madsen Itera II or GSI-61 audiometer. Air conduction thresholds were recorded for 250–8000 Hz using insert earphones (ER-3A) or supra-aural (TDH-39P) transducers. Where a hearing loss was found, bone conduction testing at 500, 1000, 2000, and 4000 Hz was performed using a Radioear B-71 bone conductor transducer to ascertain conductive or sensorineural hearing loss.

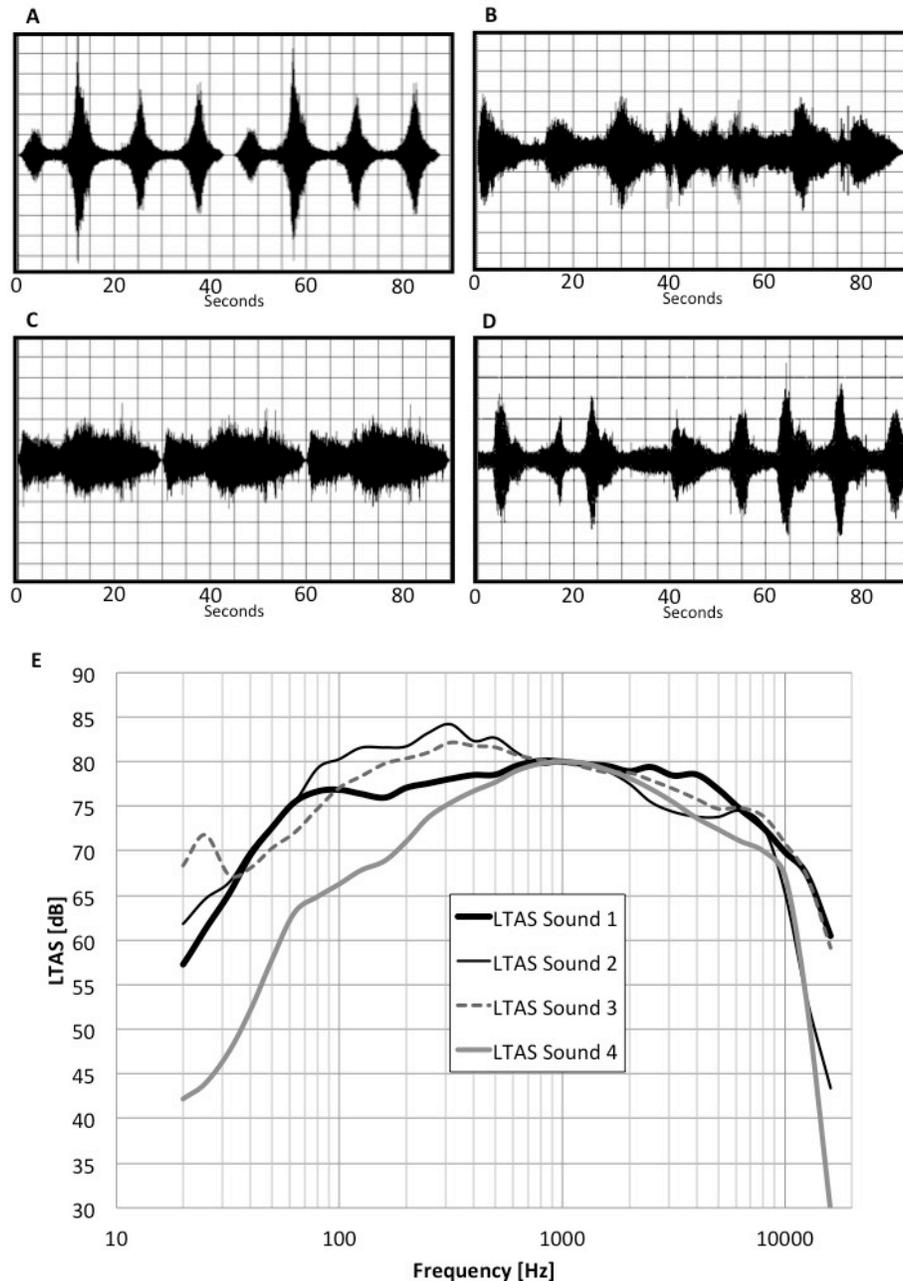
Custom Labview-based tinnitus testing software was used to determine tinnitus pitch of tinnitus with the participant responding to a 2-alternative, forced-choice method of pitch matching using tones at 15 dB SL (Sensation Level). To ensure there was no octave confusion, the matched pitch was re-presented to the participant and compared to tones that were one octave above and one octave below the estimated pitch.

Part 1: Natural Surf Sounds

Participants. Ten participants (5 men and 5 women) aged 37 to 68 years ($M = 57$ years) took part in this study. Participant characteristics are summarized in Appendix A.

Preparation of stimuli. Four audio recordings of ocean surf sounds, each 1.5 minutes in duration, were downloaded from the Internet (<https://freesound.org/>). The recordings

Figure 1



Temporal waveforms (envelopes, normalized scale); and E. Spectra of each nature sound recording (A–D). Visual analysis of the temporal waveforms of the four sound stimuli show that the various waves making up Sounds B and C had a quick rise and relatively slow decay (described as damped). Sounds A and D, on the other hand, had a combination of damped and ramped (gradual rise and relatively quick decay) waves. The waves of Sound A were also more symmetrical in shape. The length of each wave varied between sounds, with Sounds A and D having waves approximately 10 seconds in length, and Sound C having waves ranging between 10 and 20 seconds. The length of the waves in Sound B varied. Sounds B and D both had irregular patterns in comparison to Sounds A and C, with the distance between peaks varying throughout the two recordings. The interval between waves in each sound stimulus also differed, with Sound A having the longest intervals, and Sounds B and C having fewer and comparatively shorter intervals. Sounds A, B, and C have similar spectra. Sound D had less energy than the other three sounds, especially at low (< 1000 Hz) and high (> 10000 Hz) frequencies.

were selected based on their subjective sound quality and differing oscillation patterns. The recordings were normalized to a consistent root mean square using Audition audio editing software (Adobe) to equalize the sound energy of all four sounds. Individual attenuation parameters obtained from the participants' audiograms (Appendix A), at all frequencies, were applied to the four nature sound recordings to compensate for any hearing loss. To see whether there were any large differences in the long-term average spectrum of the four sound stimuli, each sound stimulus was measured by analysis of the average sound energy in each 1/3 octave band (**Figure 1B**).

Participants were asked to listen to the initial four 1.5-minute segments of ocean surf sound recordings played via Labview software (National Instruments, Austin, TX) on a computer connected to Sennheiser HD-280 pro circumaural headphones (Sennheiser, Germany) to determine their desired level for tinnitus masking. A method of adjustment was used with the researcher presenting the sound in 2 dB steps. Participants were instructed to select their desired level of sound masking based on comfort and reduced audibility of tinnitus at the time of testing. The desired level was described as "the lowest level that provided tinnitus relief and was comfortable to listen to for 5 minutes." The desired level did not vary greatly between sounds (standard deviation 1.6 dB). The test level was the average sensation level across the four sounds (Appendix A). Participants then listened to each sound at their chosen level for 5 minutes without making judgements. The stimuli were presented in counterbalanced order.

Rating scales. Following the 5-minute presentation of each sound, participants were asked to complete three rating scales while continuing to listen to the sound. A tinnitus loudness scale and tinnitus annoyance scale were used to evaluate the participants' subjective ratings of tinnitus loudness and tinnitus annoyance, respectively, in response to each sound. The tinnitus loudness scale and tinnitus annoyance scale ranged from the extremes of *not annoying at all* (1) to *very annoying* (10) and *not loud at all* (1) to *very loud* (10). The stimulus annoyance scale was used to evaluate participants' subjective ratings of stimulus annoyance and ranged from *very annoying* (1) to *pleasant* (10).

Round-robin tournament. A round-robin tournament (balanced paired-comparison method) of 6 trials was then used in which each of the four sounds were compared against every other sound to determine which sound was the most preferred across the comparisons (sound A vs. B, A vs. C, A vs. D, B vs. C, etc.). Each sound was played for as long as participants needed to make their judgement.

Preferences between sounds were recorded and the standard ranking ("1, 2, 2, 4") method was used to order the sounds according to individual preference and account for ties.

Part 2: Simulated Sounds

Participants. A total of 10 participants (7 men and 3 women) aged 22 to 68 years ($M = 54.2$ years) took part. Five participants from Part 1 also participated in Part 2. Participant characteristics are summarized in Appendix A.

Preparation of stimuli. A pink noise track was generated using the Audition software. The pink noise was presented to the participants and the threshold level was obtained using an ascending method in 2 dB steps. Participants were instructed to select their preferred level of sound masking based on comfort and reduced audibility of tinnitus as described in Part 1. This level was used to test all temporal varieties of the noise (Appendix B). Labview software was used to generate nine simulated surf sound tinnitus maskers from the pink noise. The duration of each of the rise and decay (gain) components of the nine simulated surf sounds was varied (**Table 1, Figure 2**). All sounds included a 2-second period of maximum intensity (plateau) and a 2-second interval between waveforms.

Round-robin tournament. A round-robin tournament of 36 trials in which each sound stimulus was compared against every other sound stimulus (paired comparison) was used to determine which sound was the most preferred. The standard ranking method was used to order the sounds according to individual preference and account for ties.

Analysis. Nonparametric analysis of results was undertaken using GraphPad Prism version 5 for MacOSX. Continuous data were evaluated using Friedman's test with post hoc analysis using Dunn's multiple comparison test. Spearman rank correlations were calculated to evaluate the relationship between individuals' selections of sounds.

Results

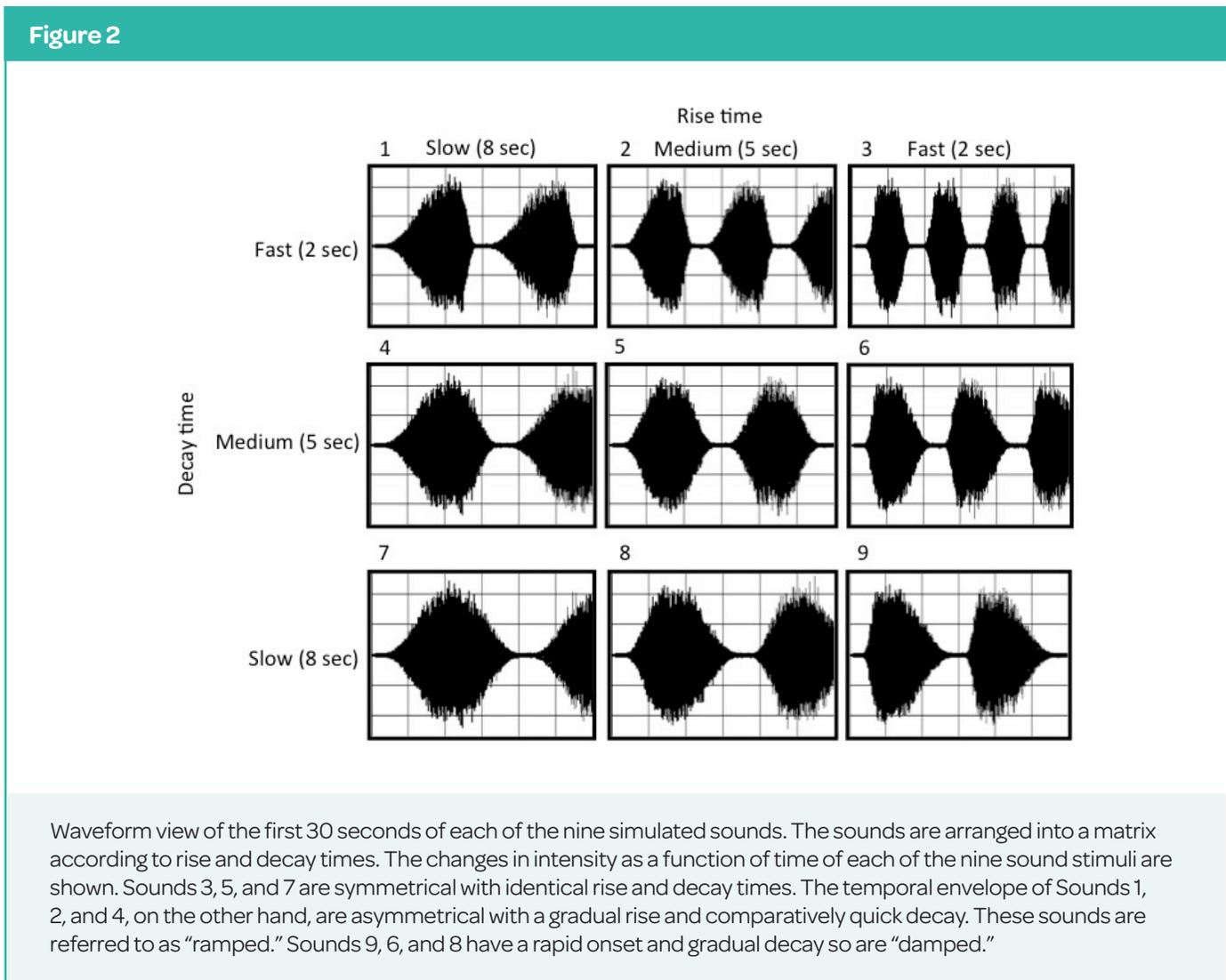
Part 1

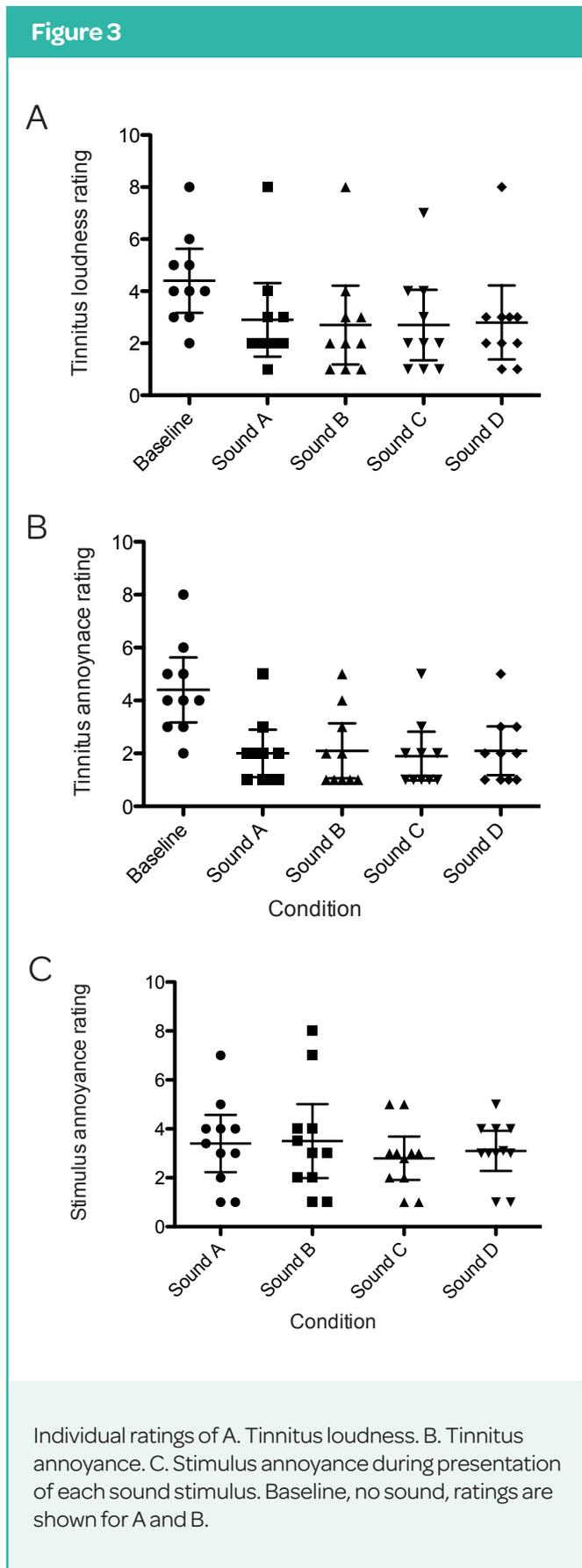
There was a statistically significant difference in tinnitus loudness depending on condition (**Figure 3A**), $\chi^2(5) = 15.1, p < .005$. Post hoc analysis using Dunn's multiple comparison test did not identify any statistically significant differences between pairs of conditions. There was a statistically significant difference in tinnitus annoyance depending on condition (**Figure 3B**), $\chi^2(5) = 29.7, p < .001$. Post hoc analysis using Dunn's multiple comparison test found all

Table 1

Temporal Characteristics of the Nine Simulated Surf Sounds Generated

	Rise (sec)	Plateau (sec)	Fall (sec)	Interval (sec)
Sound 1	8	2	2	2
Sound 2	5	2	2	2
Sound 3	2	2	2	2
Sound 4	8	2	5	2
Sound 5	5	2	5	2
Sound 6	2	2	5	2
Sound 7	8	2	8	2
Sound 8	5	2	8	2
Sound 9	2	2	8	2





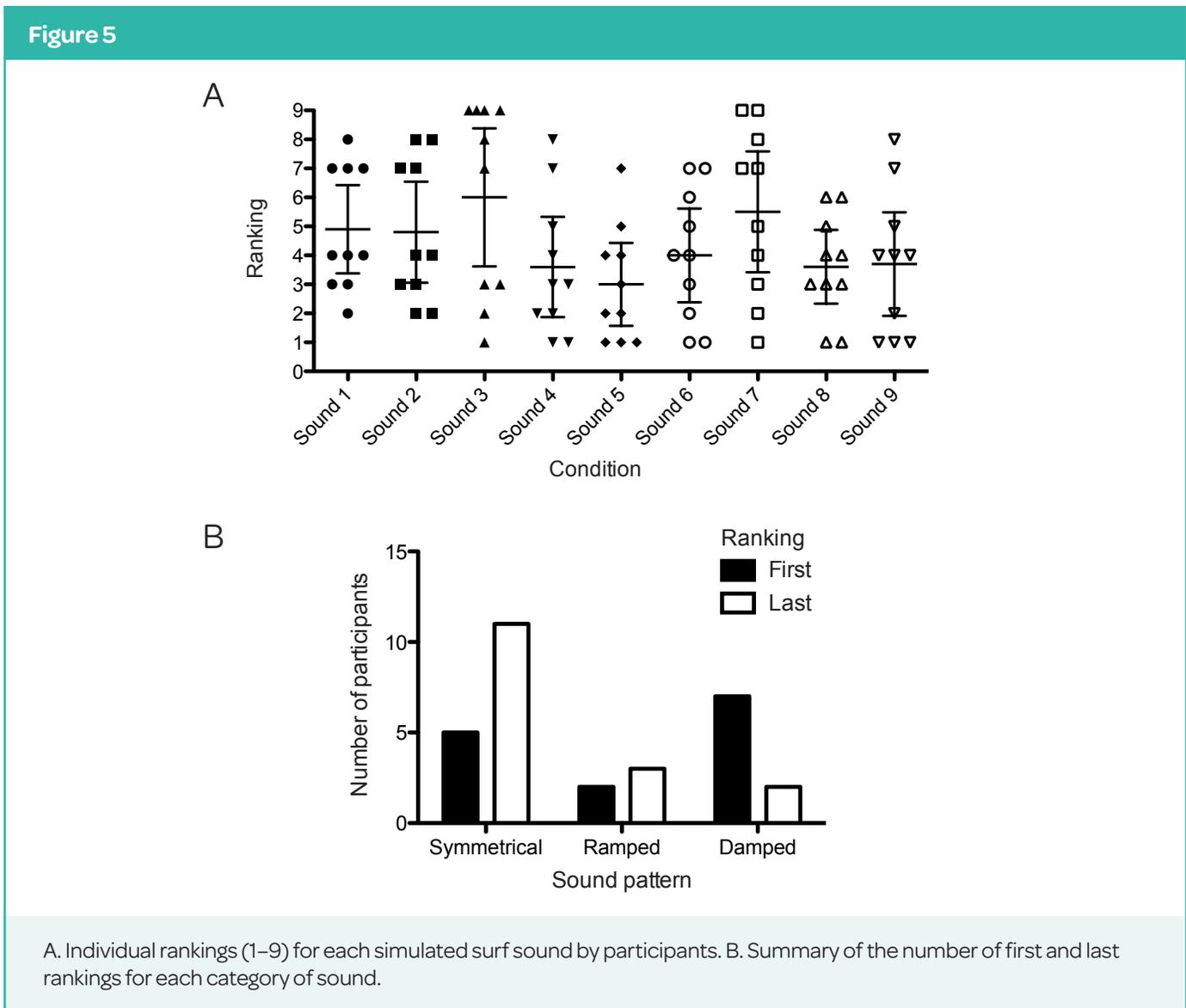
sounds had lower median annoyance ratings compared to baseline (baseline median = 4.0; Sound A median = 2.0, $p < .05$; Sound B median = 1.5, $p < .05$; Sound C median = 1.5, $p < .01$; Sound D median = 2.0, $p < .05$). However, there was no statistically significant difference in stimulus annoyance between the sound conditions (Figure 3C).

Standard rankings for the round-robin tournament (Figure 4A) showed that Sound C had the lowest (best) ranking (median = 1.0) compared to Sound B and Sound D (median ranking = 2.5) and Sound A (median ranking = 3.0). A Spearman's rank-order correlation was run to determine the relationship between the sound rankings (matrix shown in Appendix B). There was a strong, negative correlation between Sound A and D, which was statistically significant, $r_s(2) = -0.68, p < .05$ (matrix shown in Appendix C). Sound C was most frequently ranked as the best sound, while Sound D was most often ranked last (Figure 4B).

Part 2

Standard rankings for the round-robin tournament (Figure 5) showed that Sound 5 had the lowest (best) ranking (median = 2.5) compared to Sound 3 that had the highest (worst) ranking (7.5). A Spearman's rank-order correlation was run to determine the relationship between the sound rankings (Appendix C). There was a strong, negative correlation between Sound 1 and 3, $r_s(7) = -0.65, p < .05$; Sound 1 and 6, $r_s(7) = -0.67, p < .05$; and Sound 1 and 9, $r_s(7) = -0.77, p < .05$, which were statistically significant. There was a strong, statistically significant positive correlation between Sound 1 and 4, $r_s(7) = 0.65, p < .05$. There was a strong, statistically significant, negative correlation between Sound 2 and 6, $r_s(7) = -0.7, p < .05$. There was a very strong, negative correlation between Sound 3 and 4, $r_s(7) = -0.86, p < .01$, and Sound 3 and 7, $r_s(7) = -0.86, p < .05$. Sound 3 had a strong positive correlation with Sound 9, $r_s(7) = 0.78, p < .05$. Sound 4 had a strong positive correlation with Sound 7, $r_s(7) = 0.76, p < .05$, and a very strong, negative correlation with Sound 9, $r_s(7) = -0.86, p < .01$. There was a strong, negative correlation between Sound 5 and 8, $r_s(7) = -0.68, p < .05$, and a very strong, negative correlation between Sound 7 and 9, $r_s(7) = -0.82, p < .01$.

The nine sounds were collapsed into three categories of symmetrical, ramped, and damped (Figure 5B). Damped sounds were slightly more frequently ranked as best (7 times) compared to symmetrical sounds (5 times) with ramped sound favoured by only two participants. The symmetrical sounds were most frequently rated last (11 times).



ramped. Although symmetrical and damped sounds were most frequently preferred, some persons preferred the ramped sounds. The correlations provide some preliminary evidence that the timing of sounds may be contributing to preference, but it is possible that there were other characteristics contributing to preferences.

The trend for damped sound over ramped sound seen in this experiment could be due to the comparatively reduced arousal evoked by damped sounds as these sounds may be perceived as moving away from the listener. Although not directly assessed here, it is possible that ramped sounds did capture attention more than damped sounds, but most, although not all, participants chose comfort over attention diversion. As the sounds elicit different activity in particular neural networks and specific cell types they may also act directly on neural assemblies that contribute to tinnitus

perception (Olsen & Stevens, 2013; Wang, Qin, Chimoto, Tazunoki, & Sato, 2014).

Conclusion

This is a proof-of-concept study that evaluated participant preferences to natural and simulated surf sounds of varying oscillatory patterns. The study did not intend to evaluate all potential combinations of oscillations nor did it purport to determine the long-term effects of preferences. The results suggest the temporal pattern of sounds be considered alongside frequency content and overall intensity as the basis for sound selection for tinnitus sound therapy. As hypothesized, there was an asymmetric response to ramped and damped sounds in reference to tinnitus perception. A slow oscillation rate also appears to be an important factor in participant preferences.

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Disclosures

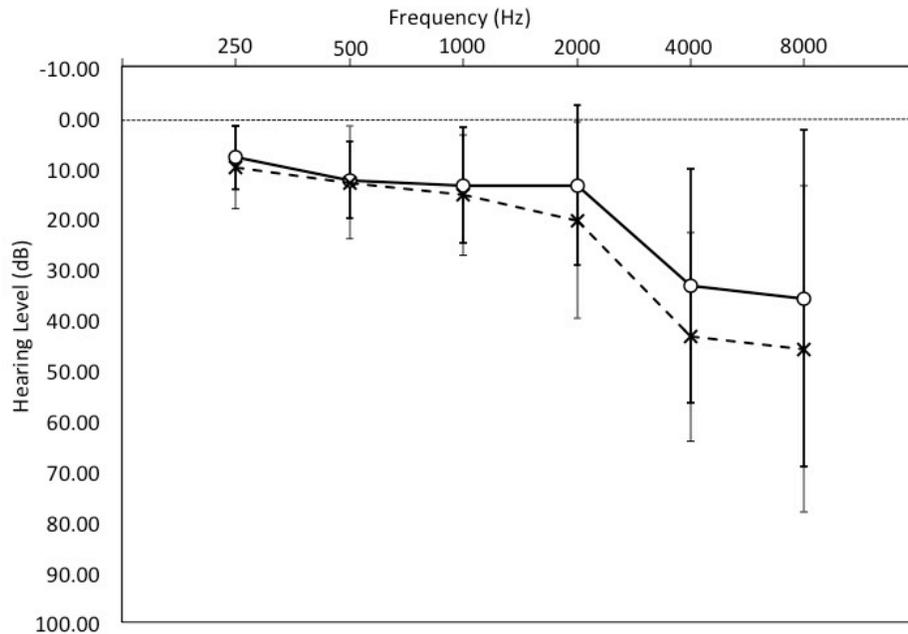
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Appendix A
Participant Characteristics Part 1

Figure A1



Average hearing thresholds of participants in Part 1, error bars represent +/- 1 standard deviation (o right ear, x left ear).

Table A1

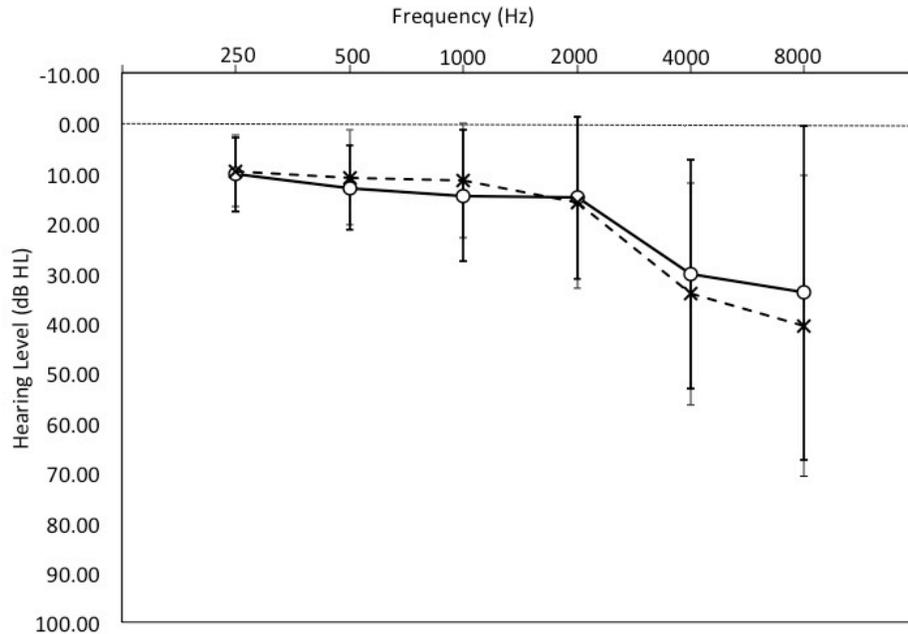
Tinnitus Characteristics of Participants in the Natural Surf Sound Comparison

	Age (years)	TFI score	Pitch (Hz)	Test level (dB SL)
1	37	53.6	12700	22
2	50	21.6	15200	20
3	61	9.6	12700	20
4	60	45.6	7900	20
5	62	6.8	6300	20
6	64	11.2	13700	28
7	68	24.0	10100	20
8	64	22.8	7000	25
9	43	18.4	9000	35
10	61	36.1	11900	28
Mean	57	25.0	10650	23.8
SD	10.2	15.6	3033.2	5.1

Note. The test level is the level at which all the sounds were presented for that individual. TFI = Tinnitus Functional Index, dB SL = decibels sensation level, SD = standard deviation.

Appendix B
Participant Characteristics Part 2

Figure B1



Average hearing thresholds of participants in Part 2 error bars represent +/- 1 standard deviation (o right ear, x left ear).

Table B1

Tinnitus Characteristics of Simulated Sound Participants

	Age (years)	TFI score	Pitch (Hz)	Test level (dB SL)
1	50	21.6	15200	29
2	68	24.0	10100	12
3	62	6.8	6300	30
4	51	24.4	10700	16
5	50	89.2	3800	26
6	58	24.0	11000	18
7	60	45.6	7900	16
8	61	36.1	11900	30
9	60	10.4	10000	33
10	22	24.4	5700	19
Mean	54.2	30.7	9260	22.9
SD	4.0	7.4	1036	7.8

Note. The test level is the level at which all the sounds were presented for that individual. TFI = Tinnitus Functional Index, dB SL = decibels sensation level, SD = standard deviation.

Appendix C
Correlation Matrices

Table C1

Correlation Matrix of Rankings for the Surf Sounds

	Sound A	Sound B	Sound C	Sound D
Sound A	-	0.22	0.45	-0.68*
Sound B	0.22	-	-0.36	-0.31
Sound C	0.45	-0.36	-	-0.30
Sound D	-0.68*	-0.31	-0.30	-

Note. * $p < .05$.

Table C2

Correlation Matrix of Rankings for the Surf Sounds

	Sound 1	Sound 2	Sound 3	Sound 4	Sound 5	Sound 6	Sound 7	Sound 8	Sound 9
Sound 1	-	0.25	-0.65*	0.76*	-0.03	-0.67*	0.65*	0.28	-0.77*
Sound 2	0.25	-	0.20	0.00	0.20	-0.70*	-0.23	-0.30	-0.23
Sound 3	-0.65*	0.20	-	-0.86**	-0.20	0.13	-0.86**	-0.28	0.78*
Sound 4	0.76*	0.00	-0.86**	-	0.39	-0.46	0.76*	-0.01	-0.86*
Sound 5	-0.03	0.20	-0.20	0.39	-	-0.24	0.34	-0.68*	-0.52
Sound 6	-0.67*	-0.70*	0.13	-0.46	-0.24	-	-0.19	0.05	0.53
Sound 7	0.65*	-0.23	-0.86**	0.76*	0.34	-0.19	-	0.12	-0.82**
Sound 8	0.28	-0.30	-0.28	-0.01	-0.68*	0.05	0.12	-	0.03
Sound 9	-0.77*	-0.23	0.78*	-0.86**	-0.52	0.53	-0.82**	0.03	-

Note. * $p < .05$, ** $p < .01$.



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