

# Service Preferences of Parents of Children With Mild Bilateral or Unilateral Hearing Loss: A Conjoint Analysis Study

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**Objective:** Universal newborn hearing screening results in substantially more children with mild bilateral and unilateral hearing loss identified in the early years of life. While intervention services for children with moderate loss and greater are generally well-established, considerable uncertainty and variation surrounds the need for intervention services for children with milder losses. This study was undertaken with parents of young children with permanent mild bilateral and unilateral hearing loss to examine their preferences for characteristics associated with intervention services.

**Design:** Conjoint analysis, a preference-based technique, was employed to study parents' strength of preferences. Using a cross-sectional survey that consisted of eight hypothetical clinic scenarios, we invited parents to make a discrete choice (to select one of two or more different services) between available services with different characteristics. The survey was informed by qualitative interviews conducted for this purpose. The questionnaire was administered to parents receiving intervention services in the province of Ontario, Canada, who were enrolled in a mixed-methods longitudinal study examining outcomes in early-identified children with mild bilateral/unilateral hearing loss. Data were analyzed using a generalized linear model (probit link) to identify attributes of interest for the respondents. Characteristics of the children were entered into the model to control for differences in age of diagnosis, sex, laterality of hearing loss, and hearing aid use.

**Results:** A total of 51 of 62 invited parents completed the questionnaire. All four attributes of care that were included in the survey were found to be statistically significant, that is, parents valued support for amplification, support for speech-language development, emotional support, and communication from professionals. Analysis showed greater preference for enhanced levels relating to support for speech-language development than for support for amplification. Preference for attributes relating to emotional support and communication were also greater than for support for amplification use.

**Conclusions:** Conjoint analysis was used to quantify parents' preferences for service attributes. Parents' values provide insights into the aspects of a service model that should receive consideration in the development of intervention programs for young children with mild bilateral or unilateral hearing loss and their families. Although parents of young children with mild bilateral or unilateral hearing loss valued several components of care, they indicated a clear preference for speech-language support compared with support for amplification use.

**Key words:** Children, Conjoint analysis, Discrete choice experiment, Hearing loss, Qualitative research, Questionnaire.

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## INTRODUCTION

There is heightened interest in the service needs of children with mild bilateral and unilateral hearing loss (McKay et al. 2008; Tharpe 2008; Fitzpatrick et al. 2016; Porter et al. 2016). The interest stems from several realities: (1) The prevalence of young children first presenting with mild bilateral or unilateral hearing loss is higher than was previously known before the implementation of widespread universal newborn hearing screening (UNHS). These children accounted for more than 40% of those identified with permanent hearing loss in one population-based study (Fitzpatrick et al. 2014); (2) children with these hearing profiles can be identified at substantially younger ages in the context of newborn screening (Fitzpatrick et al. 2014, 2017a; Ghogomu et al. 2014) even when not specifically targeted as part of the disorder (Wood et al. 2015); (3) the longer term impact of early identified mild bilateral or unilateral loss remains relatively unknown. One reason information continues to be limited is that not all children with mild bilateral or unilateral loss are specifically targeted through newborn screening programs due to the uncertainty around the advantages of early intervention (Wood et al. 2015). In addition, a report from Johnson et al. (2005) concluded that the majority of infants missed by screening programs have mild hearing loss. As stated by the Centers for Disease Control and Prevention (2005), children with mild loss may be missed because screening procedures are not sensitive to these degrees of hearing loss.

Studies from the 1980s and 1990s, based primarily on children with late-identified mild bilateral/unilateral loss, showed that these children were at risk for language and academic concerns. These studies and several literature reviews have documented that children lagged behind their normal hearing peers in vocabulary, reading, and phonological abilities and that up to one third failed a grade in school (Bess et al. 1986; Bess et al. 1998; Wake & Poulakis 2004; Winiger et al. 2016). More recent studies with clinical populations continue to flag the need for concern especially in the school years (Lieu et al. 2010; Dokovic et al. 2014; Fischer & Lieu 2014; Porter et al. 2016). However, one population-level study in Australia did not find differences in outcomes, with the exception of phonological memory skills, in school-age children with slight/mild hearing loss compared with normal-hearing peers (Wake et al. 2006). It is noteworthy that the majority (69%) of the 48 children assessed had hearing loss in the "slight" range (16 to 25 dB HL).

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Even when hearing loss is identified early due to UNHS, most studies point to poorer language development than normal hearing children, both for children with mild bilateral and unilateral loss (Sedey et al. 2005; Yoshinaga-Itano et al. 2008). A recent longitudinal study found minimal impact on spoken language outcomes in early-identified children during the early preschool years (Fitzpatrick et al. 2015). However, that report on children up to age 3 years relied heavily on parent questionnaires. As reported more recently, at later preschool ages, these children performed on average at lower levels in some areas of communication development compared with the normal-hearing study control group. In particular, children with unilateral hearing loss showed gaps in receptive and expressive language skills at age 48 months (Fitzpatrick et al. 2018). In addition to the negative consequences for language development, attention to these milder losses is warranted because of the risk of progressive hearing loss. Research has shown that 20 to 30% of children with mild bilateral or unilateral hearing loss experience deterioration in hearing over time (Barreira-Nielsen et al. 2016; Fitzpatrick et al. 2017a).

There has been a shift toward earlier fitting of amplification and management of children with milder degrees of hearing loss (Holte et al. 2012; Fitzpatrick et al. 2014, 2017b; Bagatto et al. 2016). However, there remains uncertainty about the benefits of intervention or amplification with considerable variation between clinical decisions. Current guidelines recommend that amplification be considered on a case-by-case basis (McKay et al. 2008; Bagatto & Tharpe 2014; Ontario Ministry of Children and Youth Services 2014; Bagatto et al. 2016). In qualitative research, parents confirmed the lack of clarity around the benefits of amplification with variation reported in recommendations from clinics and between audiologists and otologists (Fitzpatrick et al. 2016). Furthermore, even when amplification is recommended, studies suggest substantially more nonuse and inconsistent use compared with children with more severe losses (Fitzpatrick et al. 2010; Fitzpatrick et al., Reference Note 1; Walker et al. 2013). More recently, data have emerged to substantiate the advantages of amplification for children with mild bilateral loss. Walker et al. (2015) found that amount of hearing aid use in children with mild bilateral loss was associated with better vocabulary and grammar in 5- or 7-year-old children. Of several variables, hearing aid use was the largest predictor of expressive language (morphosyntax scores), accounting for 26% of the unique variance in scores. The authors proposed that early intervention services may support more consistent hearing aid use.

Given the substantial literature that points to children with mild bilateral/unilateral loss being at risk for difficulties in language and/or academic functioning (Winiger et al. 2016; Appachi et al. 2017), coupled with the indecision or inconsistent use around amplification (Fitzpatrick et al. 2010), interest has grown in whether services should be provided and what services are of value to parents. In contrast to previous generations, in regions where mild loss is targeted as part of UNHS, a substantial number of these children will now enter early hearing intervention programs well before the child's first year (Fitzpatrick et al. 2015). While there is little debate about the need for direct intervention and parent support for children with moderate loss and greater, the situation for these milder losses is less clear. This is likely due in part to limited information about whether intervention can make a difference and what

the components of appropriate intervention should be. Given our lack of knowledge in this domain, and faced with growing numbers of early identified children with these losses, it seems reasonable to elicit information about intervention needs from families. In this study, we sought to consider what elements parents consider to be important in an intervention program for families of these young children.

However, eliciting patient preferences for the various characteristics of health care services is not straightforward. One technique for quantifying preferences that has gained traction in investigating health care questions is discrete-choice experimentation or conjoint analysis (CA) (Reed Johnson et al. 2013; Kleij et al. 2017; Vass et al. 2017). The CA approach, originally developed for consumer studies in marketing, is a survey method encompassing data collection and analysis. The underlying theory of CA is that a product (including a health care service) can be described by its characteristics or attributes (Ryan et al. 2001; Reed Johnson et al. 2013). CA provides an estimation of the relative importance people attribute to various components of care, measures how individuals are willing to trade between these characteristics (attributes), and estimates the overall satisfaction they gain from various forms of health services provision. The concept of trading refers to assessing the degree to which individuals prefer improvements in one attribute to the improvements in others. Thus, trade-offs relate to accepting a less preferred version of one attribute for improved versions of others. By taking into account the various characteristics of a service from a patient satisfaction perspective, this information can support decision-making in setting priorities for various health care services. Applications of CA to health care were popularized in the 1990s and applied to elicit patient preferences in areas such as doctor-patient relationships, outcomes, and health care services (Ryan et al. 2001).

In a previous CA study of parent preferences for services for children with hearing loss, well-coordinated clinical services with access to support from other parents emerged as important characteristics of care (Fitzpatrick et al. 2007). However, the study was conducted just a few years after the implementation of a UNHS program in the province and included children across the spectrum of hearing loss. The current research was undertaken in the context of a broader study aimed at investigating developmental outcomes in children identified early with mild bilateral or unilateral loss. The project also included a focus on parent perspectives on the early identification process and their needs following diagnosis. We used CA to examine the values parents place on the characteristics of services they receive. Specifically, through this inquiry, we sought to identify the relative importance of different attributes and levels of service provision for families of children with mild bilateral and unilateral hearing loss.

## MATERIALS AND METHODS

### Design

This inquiry involved a cross-sectional questionnaire-based CA with families of children identified with mild bilateral or unilateral hearing loss. The questionnaire was developed following established CA practices that have been widely applied in health services research (Ryan et al. 2001; Reed Johnson et al. 2013). The application of the CA methodology in this study is described in the section on Procedures below.

**Context and Participants**

The province of Ontario implemented a universal newborn hearing screening program in Ontario in 2002 with well-defined protocols for assessment and audiologic follow-up including amplification and guidelines for intervention (Hyde et al. 2004; Ontario Ministry of Children and Youth Services 2014). Children receive all audiologic and intervention services through a publicly funded health service system.

In the context of this UNHS program, a longitudinal study was initiated in 2009 to examine developmental outcomes in children with mild bilateral and unilateral hearing loss. One component of the study involved a CA to elicit parents’ preferences for services. Children and their parents from four regions in Ontario, Canada, were enrolled in the full longitudinal multi-center study. The parents of all children remaining in the study at the time the CA was conducted (n = 62) were invited to participate in this component of the research. Inclusion criteria, consistent with the larger study, were as follows: permanent mild bilateral or unilateral hearing loss; age 3 years or less at study enrolment; English spoken in the home; no disabilities that would interfere with the child’s ability to complete the auditory and spoken language test battery required for the study. For this study, mild bilateral and unilateral hearing losses were defined according to the definition proposed by the Centers for Disease Control and Prevention (2005). Mild bilateral hearing loss refers to average pure-tone air conduction thresholds (at 0.5, 1, and 2 kHz) between 20 and 40 dB HL or thresholds >25 dB HL at two or more frequencies above 2 kHz; for this study, mild bilateral loss was determined based on better ear hearing thresholds. Unilateral hearing loss refers to hearing loss in one ear only with a pure-tone average ≥20 dB HL or > 25 dB at two or more frequencies above 2 kHz.

The study received ethics approval from the Children’s Hospital of Eastern Ontario (main study site) and the University of Ottawa, as well as all participating institutions. Written informed consent was obtained from each participant.

**Procedures**

**Baseline Characteristics of Children** • As part of the larger study, parents were asked to complete an initial study intake form at enrolment to provide baseline characteristics related

to the child and family (i.e., child’s hearing health history, languages used in the home and socioeconomic status). They also completed an intervention-specific questionnaire at enrolment and at annual intervals, where they reported information on amplification recommendations and use as well as type of therapy services (e.g., auditory-verbal therapy). Audiologic information was entered and updated through collection of annual reports obtained with consent from the child’s audiologist, supplemented with information from these parent questionnaires. Hearing aid use was based on parent report from the annual questionnaire, where parents reported whether amplification was recommended (yes, no) and whether it was used (yes, no). Parents categorized amount of use on a Likert scale as all day (all waking hours), most of the time (more than 6 hours), not often (less than 6 hours), and never. This information was collapsed into four categories. Consistent use referred to amplification use all day or most of the time throughout the study; inconsistent use referred to not often (less than 6 hours use) or a rating of “never used” reported at some time during the course of the study; not used corresponded to a “never” response on the annual questionnaires throughout the study; not recommended indicated that the child had not received a recommendation for amplification.

**Development of CA Questionnaire • Establishing the Attributes** • The main steps applied in developing the CA questionnaire are outlined in Figure 1 and detailed below. Stage one of the study involved identifying the principal attributes, that is, the features or characteristics of the services for young children with hearing loss and their families. Qualitative research methods have been promoted as good practice in discrete choice experiments (Reed Johnson et al. 2013; Abihiro et al. 2014; Vass et al. 2017). Qualitative interviews previously reported (Fitzpatrick et al. 2016) constituted the main data source to inform the content of the CA questionnaire. Specifically, the findings of semistructured interviews with a subset of 20 parents were used to select the characteristics (attributes) of services identified by parents and to define the levels of care. The qualitative study comprised parents from the full study; therefore, they were also part of the study group who were invited to complete the CA questionnaire. The interview group included parents of 9 children with mild

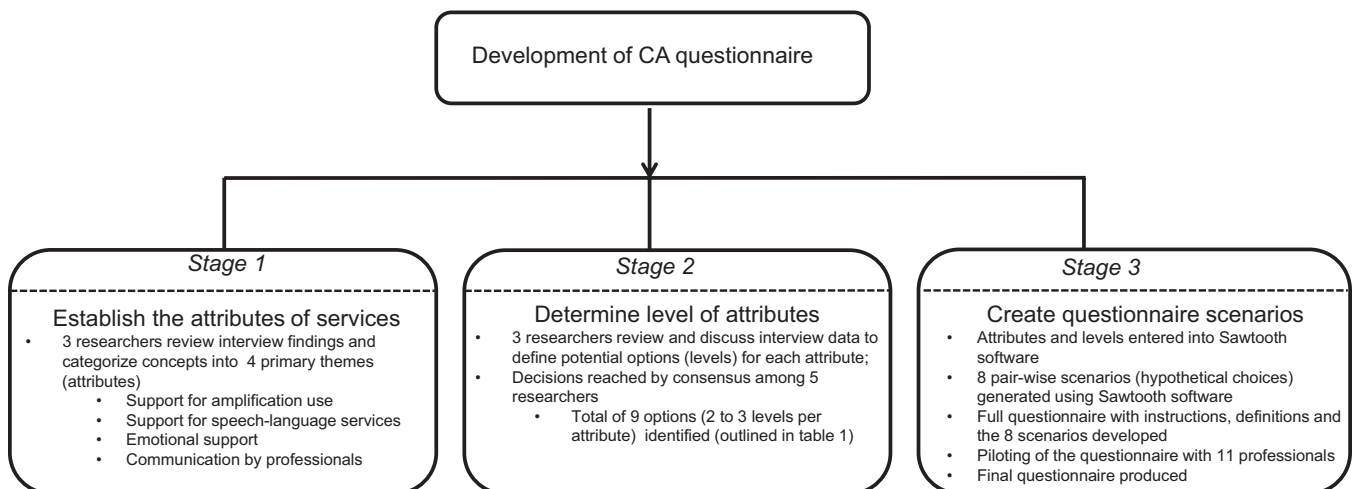


Fig. 1. Description of conjoint analysis (CA) questionnaire development.

bilateral and 11 with unilateral hearing loss from 4 different regions of Ontario. At the time of the interviews, the mean age of the children was 28.6 months (SD 9.3). Interviews were conducted with the explicit intention of acquiring information for the CA and included questions about parents' experiences around diagnosis, their needs, and preferences regarding follow-up services.

In preparing the CA questionnaire, a team of three researchers re-reviewed all concepts from the interviews and categorized them into key themes, which, after discussion, were finalized as the main attributes for the analysis. On the basis of the interviews, supplemented by literature on mild bilateral/unilateral hearing loss, four primary attributes were selected as being important to explore with families: support for amplification use, support for speech-language development, and emotional support (parent support included as part of clinic sessions with referral to other professionals when needed) and communication by professionals. Although parents identified other characteristics during the interviews, these dominated the discussion and narrowing down key attributes to four to six is necessary to design a manageable CA questionnaire.

**Level of Attributes** • In the second stage, levels (options) were then defined for each of the attributes. A level is a choice (e.g., home therapy, clinic-based therapy) that describes an attribute, and each attribute must be defined by two or more levels, so that scenarios can be created. The goal is to determine the value placed on the level by comparison to the other levels of the attribute, that is, the utility assigned to the level. One of the most difficult aspects of defining levels is identifying an appropriate spectrum of realistic choices for each attribute. In this study, attributes and their levels were finalized by reviewing and discussing the interview data until decisions were reached by consensus. The four attributes selected and their levels are provided in Table 1.

**The Questionnaire Scenarios** • The third stage involved framing questions around the attributes. In CA, because all

possible attributes and levels cannot be included in a manageable self-completion questionnaire, it is necessary to reduce the combinations to a reasonable number of scenarios (hypothetical choices). The attributes and levels were entered into Sawtooth software (SSI web version number 8.3.10, Orem, UT) to generate the final clinical scenarios that consisted of eight pairwise choices. In each pairwise choice, scenarios were identified as Service A and Service B, and their individual characteristics were presented. Essentially, through these scenarios, parents were asked to make a binary choice, that is, to choose the clinical scenario that provided them with the most satisfaction (utility) in caring for their child with hearing loss. Participants were instructed to choose the service they would select for their child's care.

The final questionnaire shown in Appendix 1 (Supplemental Digital Content 1, <http://links.lww.com/EANDH/A472>) included a section on instructions, the eight discrete choice scenarios, and definitions of each attribute and level. Before distribution, all scenarios were carefully reviewed by the research team as well as three auditory-verbal therapists for appropriateness and clarity. A final step of revision and piloting of the survey for clarity and ease of completion was carried out with 11 professionals working in pediatric hearing clinical or research services. Subsequently, minor revisions were carried out to improve clarity in wording and instructions.

The questionnaire was first distributed to parents by email followed by a total of four reminders including two email reminders, one at the end of week 1 and one at the end of week 3. A paper copy was sent to nonrespondents at the end of week 5, and finally, a link to the Sawtooth version of the questionnaire was sent to the remaining parents 4 weeks after the paper copy was sent (week 9).

**Data Analysis** • All data were analyzed using SPSS, version 24 (IBM Corp.). For the CA, a generalized linear model (probit link) was fit to identify features of interest for the respondents. Attributes were entered into the model as fixed effects in addition to a random intercept to account for the multiple responses from each individual. The respondents' ratings for each scenario formed the dependent variable. Categorical variables were transformed into dummy variables to permit regression modeling. In the regression, the coefficients (betas) with their statistical significance represent the utilities (preference scores) for the levels. This provides information about whether the attributes impact respondent choices and the relative importance of each attribute, that is, whether individuals are willing to trade (i.e., accept a less preferred version of one attribute for improved versions of other attributes). We also explored whether characteristics at the individual level related to laterality (unilateral versus bilateral), sex of child, age of diagnosis, and hearing aid use (consistent use, inconsistent use, not used, not recommended) modified parents' preferences.

## RESULTS

### Participants

A total of 51 (82.3%) of 62 parents returned the questionnaire. These children had reached a mean age of 5.2 years (SD: 1.5; range 2.2 to 8.2) when their parents completed the questionnaire. Table 2 shows the clinical characteristics of the children as well as a comparison to the full group of 62 children

**TABLE 1. Attributes and level for the conjoint analysis (CA) questionnaire**

Attribute	Level
Support for amplification	Regular visits to the clinic for the first year after the child gets hearing aids or an FM system plus the audiologist checks in with the parents regularly
	Regular visits to the clinic for the first year after the child gets hearing aids or an FM system
Support for speech-language development	Therapy sessions at the clinic or at home with the child and parent
	Regular monitoring of development through speech-language assessments
	Parent information sessions
Emotional support	Regular part of service
	Parents seek support independently
Professional communication	Warm and supportive
	Business-like

CA design  $3^2 \times 1^3$ .

Questionnaire included definitions of each attribute and level.

FM, frequency modulated system.

**TABLE 2. Characteristics of children with mild bilateral or unilateral hearing loss whose families completed conjoint analysis questionnaire**

Characteristics	Invited (n = 62)	Returned (n = 51)
Sex, n (%)		
Male	32 (51.6)	24 (47.1)
Screening status, n (%)		
Screened	57 (91.9)	47 (92.2)
Not screened or unknown status	5 (8.1)	4 (7.8)
Age at confirmation (mos), median (IQR)	3.8 (2.5–5.9)	4.0 (2.7–6.1)
Hearing loss laterality, n (%)		
Unilateral	34 (54.8)	28 (54.9)
Bilateral mild	28 (45.2)	23 (45.1)
Onset of hearing loss, n (%)		
Congenital	59 (85.5)	42 (82.4)
Early onset	4 (5.8)	4 (7.8)
Late onset	3 (4.3)	3 (5.9)
Unknown	3 (4.3)	2 (3.9)
Type of hearing loss, n (%)		
Sensorineural	48 (77.4)	39 (76.5)
Permanent conductive	14 (22.6)	12 (23.5)
Onset of hearing loss, n (%)		
Congenital	52 (83.9)	42 (82.4)
Early onset ( $\leq 6$ mos)	4 (6.5)	4 (7.8)
Late onset ( $> 6$ mos)	3 (4.8)	3 (5.9)
Unknown	3 (4.8)	2 (3.9)
Etiology, n (%)		
Neonatal intensive care unit	1 (1.6)	0
Cytomegalovirus	2 (3.2)	2 (3.9)
Syndromes	3 (4.8)	3 (5.9)
Hereditary/genetic	8 (12.9)	8 (15.7)
ENT malformations	12 (19.4)	10 (19.6)
Etiology unknown	36 (58.1)	28 (54.9)
Age recommendation for amplification (mos), median (IQR)*	6.4 (3.5–21.6)	7.4 (3.6–23.2)
Age fitting of amplification (mos), median (IQR)†	11.2 (5.6–22.8)	11.3 (5.6–22.8)
Hearing aid use, n (%)		
Consistent use	35 (56.5)	30 (58.8)
Inconsistent use	7 (11.3)	6 (11.8)
Not used	11 (17.7)	8 (15.7)
Not recommended	9 (14.5)	7 (13.7)

\*Age at recommendation of amplification was available for 50 of 53 children in invited group who received amplification recommendations and for 44 of 44 children in conjoint analysis group.

†Age of fitting was available for 42 of 48 children fitted with amplification in invited group and for 38 of 42 children in the conjoint analysis group.

ENT, ear nose throat (anomalies included atresia and microtia); IQR, interquartile range.

whose parents were invited to participate. As shown, the characteristics of the children seemed to reflect those of the full group. Consistent with our regions of recruitment, children came from four different regions in Ontario. The majority (92.2%) were exposed to newborn screening, resulting in an early identified group of children with a median age of diagnosis of 4.0 months (interquartile range: 2.7 to 6.1). Just over half (54.9%) had unilateral loss and 45.1% had mild bilateral loss at initial diagnosis, consistent with our full study sample. A total of 90.2% (n = 46) had congenital or early-onset loss (onset < age 6 months), and 76.5% had sensorineural loss with the remaining having permanent conductive disorders (23.5%).

Amplification was recommended for 44 (86.3%) of the children and 42 (82.4%) of them had been fitted at a median age of 11.3 months (interquartile range: 5.6 to 22.8). As noted, hearing aid use (Table 2) was based on parent report from a questionnaire completed annually. As shown in Table 2, 35 (56.5%) of all children used amplification consistently. Parents all reported receiving some level of intervention at some point during the preschool years, but the frequency and type of provider varied (e.g., auditory-verbal therapist, teacher of the deaf and hard of hearing, speech-language pathologist).

### CA Questionnaire Findings

As noted, a generalized linear model (probit link) was fit to identify the attributes of interest for the respondents. We also conducted a further analysis controlling for the following variables: laterality (unilateral, bilateral), sex of child, age of diagnosis, and hearing aid use (four categories described above), but the results were unchanged. Table 3 presents both the unadjusted and adjusted results obtained from the analysis.

As shown in Table 3, the coefficients for all attributes in the regression model are significant at the  $p < 0.01$  level. In summary, the results indicate that all four attributes: support for amplification use, support for speech-language development, emotional support, and professional communication were valued as characteristics of a service model by these respondents. In this model, the relative strength of preference of respondents between attributes is expressed by the ratio of the regression coefficients. Specifically, the results for each of the four attributes presented to parents show that they preferred (1) regular visits to the audiology clinic plus audiologist check-in over regular visits to audiology only, that is, parents were 1.4 times more likely to prefer support with amplification use with outside clinic check-ins over regular visits only; (2) regular therapy sessions at the clinic over information sessions only and regular monitoring of development through speech-language assessments over parent information sessions only; parents were 2.77 times more likely to prefer therapy sessions at the clinic over information sessions only; (3) emotional support to be part of regular service rather than having to seek it out independently; and (4) professional communication to be warm and supportive rather than business-like.

The results show a clear preference from these parents for enhanced level of support for speech language development over enhanced level of support for amplification use. For example, as shown in Table 3, the odds ratios for support for speech-language development show that the odds of a parent preferring therapy sessions at the clinic or regular language monitoring was twice (2.77/1.40; 2.81/1.40) that for enhanced support for amplification use. However, care should be taken in the interpretation of further such calculations as the study is not powered to detect small differences in the magnitude of odds ratios.

### DISCUSSION

This study used CA techniques to obtain preferences for health services from parents of children with mild bilateral and unilateral hearing loss. Our results, coupled with the qualitative interview data that informed the questionnaire, indicate that parents value the four attributes of services that were presented to them (in the questionnaire) as characteristics of a service model. These included support for amplification use, support

**TABLE 3. Results of conjoint analysis—random-effect probit model (n = 51)**

Attributes	B (95% CI)	Exp (B) (95% CI)	p	B adjusted (95% CI)	Exp (B adjusted) (95% CI)	p
Support for amplification use (ref: regular visits only)	0.33 (0.14–0.53)	1.40 (1.15–1.70)	0.001	0.34 (0.14–0.54)	1.40 (1.15–1.70)	0.001
Support for speech-language development (ref: parent information sessions)						
Therapy sessions at the clinic	1.02 (0.76–1.28)	2.77 (2.13–3.60)	<0.001	1.00 (0.74–1.27)	2.72 (2.09–3.55)	<0.001
Regular monitoring	1.03 (0.80–1.27)	2.81 (2.22–3.56)	<0.001	1.01 (0.78–1.25)	2.75 (2.17–3.49)	<0.001
Emotional support (ref: parents seek support independently)	0.75 (0.56–0.95)	2.12 (1.74–2.58)	<0.001	0.74 (0.54–0.94)	2.09 (1.72–2.55)	<0.001
Professional communication (ref: business-like)	0.74 (0.52–0.95)	2.09 (1.69–2.58)	<0.001	0.72 (0.51–0.93)	2.05 (1.66–2.55)	<0.001

*“Reference” refers to reference level for each attribute.*

*Support for speech-language development has three levels; all other attributes have two levels.*

*Exp (B) refers to the utilities (preference scores) for the levels.*

*CI, confidence interval; Ref, reference level for each attribute.*

for speech-language development, emotional support, and communication from professionals. Although these parents valued all of these attributes, they showed a preference for enhanced support for speech-language development over other attributes of clinical support services. In fact, this study showed the odds of a parent preferring support for language development was twice that for support for amplification use. For amplification support, parents preferred a model that includes regular clinical audiology visits and regular check-ins (phone calls or email), over clinic visits only. For language development support, they preferred therapy visits at a clinic to information sessions only and regular monitoring of language development to information sessions only. They showed a preference for emotional support as part of the clinical service and for warm and friendly professional communication. After controlling for child characteristics, there was no difference in results. The relationship between the preferences and the attributes of service was not affected by the child’s characteristics (mild bilateral versus unilateral hearing loss, sex, age of diagnosis, amount of hearing aid use).

Despite their children’s milder degree of hearing loss, these parents seemed to prefer a preventative model for language development support that comprised ongoing direct services. This was consistent with our findings from the interviews where parents spoke of their appreciation for the support they received through direct services with specialists. This gave them confidence that language was on track or progressing (Fitzpatrick et al. 2016). However, in our questionnaire, we did not probe frequency or dose of direct intervention.

Although there is good consensus that children with hearing loss require intervention services (Joint Committee on Infant Hearing 2007; Moeller et al. 2013), the situation is not entirely clear for children with mild bilateral or unilateral hearing loss. For example, in the United States, researchers have reported that children with mild bilateral loss do not meet eligibility criteria for early intervention services in all states (Holstrum et al. 2008) or even for intervention at school age (Yoshinaga-Itano et al. 2008). In Ontario, where this study was conducted, parent information collected through our study intake questionnaires and through our parent interviews suggests that availability of, intensity, and type of services for these children vary from region to region (Fitzpatrick et al. 2016). As noted above, one of the most important issues emerging from our parent interviews was around hearing aid use in the early years. It is

possible that parents also see direct therapy as providing support with this part of the care. By definition, CA breaks down a service into discrete components. It is quite possible that we did not adequately separate the differences between audiologic and intervention services. Certainly, from parents’ perspectives, consistent care is needed regardless of degree of hearing loss. This notion has emerged in other interviews where parents of children across the full spectrum of bilateral hearing loss valued coordinated therapy services and access to information (Fitzpatrick et al. 2008).

One of the primary issues parents raised during the interviews that informed this CA was the uncertainty around the benefits of hearing aids and the challenges in achieving consistent use with their young children. One finding of interest related to support for amplification use in the early stages of care was that parents preferred regular audiology visits combined with regular check-ins by their audiologist (defined as 1- to 2-month intervals) outside the onsite clinical visits. The challenges of consistent hearing aid use for young children have been well highlighted in the literature (Walker et al. 2013; Muñoz & Hill 2015), and milder degree of hearing loss is one of the factors affecting amount of use.

In a previous CA study (Fitzpatrick et al. 2007) that investigated parent preferences for care related to children with hearing loss, coordinated care dominated the model, as the most important attribute for families compared with other service components. However, in our qualitative interviews that informed the current study (Fitzpatrick et al. 2016), parents did not identify coordination of services as an important component of service, and, therefore, it did not appear in the attributes presented in this questionnaire. One key difference between the two studies is that the 2007 study included children across the range of hearing loss, with the majority having severe to profound loss. Parents’ needs and what they consider to be most important in a package of care may be different when hearing loss is less severe. It is also important to note that the previous study took place in Ontario shortly after implementation of the province-wide UNHS program and that some children had been diagnosed before or at the start of the program. The Ontario IHP program, implemented since 2002 (Hyde et al. 2004), provides protocols for screening, assessment, and intervention. The children in the current study were all identified since the screening program was initiated. Although we cannot draw a definitive

conclusion from this study, it is possible that services are now perceived as better coordinated. This may reflect the maturity of the newborn hearing screening services and the fact that services are well-organized.

An important principle of CA is that the attributes, and their levels should reflect the characteristics of the service, in that they are meaningful and realistic to potential users. The qualitative interview data from the same population (Fitzpatrick et al. 2016), where questions of service needs were specifically explored, helped strengthen the content of this CA questionnaire. Testing and reading of the survey by professionals in related research and clinical aspects before distribution assisted with clarity and ease of completion.

### Limitations

An inherent limitation of a CA questionnaire is that it is restricted to a set number of profiles, from which respondents can choose. We selected the attributes that were judged to be most important to parents. However, it is possible that we did not capture the nuances, for example, between speech-language services and audiologic services in providing support for amplification use. As noted by Walker et al (2015) in their study on children with mild hearing loss, amplification use is closely linked to intervention, and it is very difficult to sort out how these two factors individually affect communication outcomes. Another limitation of this study, described in other CA studies (e.g., Viney et al. 2002), was that respondents can only rank components of models that they have experienced or that they can envision. This may, in part, explain some of the parents' preferences. In our interviews, they described what they knew, and these items constituted the main foundation of the questionnaire. Our study was also limited to one Canadian province, although it had the advantage of including participants from four different regions with some variations in intervention services. Finally, despite attempts to recruit a broad range of families, the participants were primarily university educated. Families from different backgrounds might have different preferences for care. Judgment about the relative importance of certain characteristics such as direct attendance at clinics for audiology and therapy might be somewhat different from families with more limited resources. It is also important to note that this study was conducted in the context of a publicly funded screening and intervention program with services provided through health and/or education programs. The findings may not reflect preferences in systems that involve user fees.

### Clinical Implications

In some jurisdictions, mild bilateral and unilateral hearing losses are not specifically targeted due to the lack of evidence related to the advantages of early identification and intervention (Wood et al. 2015). This question remains largely unanswered. However, in regions where these children are being identified early due to UNHS (Fitzpatrick et al. 2014), the issue of whether to provide them with early intervention services and the essential components of an appropriate service model have become of interest. CA is increasingly used as an approach to inform decision-making across an array of health care concerns (Reed Johnson et al. 2013). This study represents an application of the method to study preferences for services from parents of children with permanent mild bilateral/unilateral hearing

loss. This research expands previous efforts through qualitative research to highlight aspects of early care that are important to families by focusing on their priorities for service components in a quantitative manner.

### Conclusions

This study provides information on the values that patients place on various components of service provision for children with mild bilateral/unilateral loss. Information on the utility (benefits) patients associate with various service components has implications for program planning. There are two key findings that have clinical importance from this study. The first is that these parents prefer ongoing on-site audiologic service that includes regular check-ins by an audiologist in the early years. Second, they also prefer direct therapy support and monitoring through assessments rather than only parent information sessions about language development. In addition, not surprisingly, parents value emotional support as part of the care package along with warm and supportive communication from professionals. An important next step would be the inclusion of the perspectives of practitioners who provide services for these children within the context of a UNHS program that must allocate services to all children with hearing loss and their families. In addition, information through CA or other techniques from a broader range of families would allow an examination of factors shown to impact preference outcomes (e.g., education and geographical location) and would contribute further insights about the components of service models for these children.

Early identification of mild bilateral/unilateral hearing loss is still a relatively recent phenomenon since the widespread implementation of population-based screening. In summary, this study provides insights into the importance of various attributes of a service model for children with mild bilateral/unilateral hearing loss from parents' perspectives. The information gleaned from this study can be useful for clinical programs when designing intervention services following early identification of mild/unilateral hearing loss. Our principal aim in undertaking this CA questionnaire combined with the previous qualitative research (Fitzpatrick et al. 2016) was to identify the key components that parents valued in a care system. Compared with children with more severe degrees of hearing loss, there has been an unsystematic approach to intervention services for these children with milder losses. This is largely because of the lack of certainty about the consequences on children's development. Even when eligibility for services per se is not an issue such as in the Ontario system, there do not seem to be clear guidelines in place. Our previous interviews with parents from multiple areas suggest there is inconsistency in services (Fitzpatrick et al. 2016). Our results from this CA study lead us to conclude that families of both children with mild bilateral and unilateral loss should be offered services that consist of direct speech-language support services, audiological follow-up in the early months including check-ins with parents outside clinic visits, and emotional support as part of the intervention program. Coupled with previous findings that a substantial proportion of children with these losses progress to bilateral or more severe hearing loss (Fitzpatrick et al. 2014, 2017), this study also has implications for decision makers as it suggests that more direct care should be included in the planning of clinical services for these children.

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E.M.F. designed the study, guided the questionnaire development, and wrote the first draft of the manuscript; D.C. assisted with the questionnaire an provided interpretive analysis and critical revision. I.G. carried out the statistical analyses and provided interpretive analysis. A.D.-S. provided guidance in the study design and critical revision. J.W. managed data collection and descriptive statistical analyses. V.G., E.N., and M.S. managed the development and piloting of the questionnaire. All authors commented on the final manuscript.

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