Empirical Article

Relationships Between Spoken Word and Sign Processing in Children With Cochlear Implants

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The effect of using signed communication on the spoken language development of deaf children with a cochlear implant (CI) is much debated. We report on two studies that investigated relationships between spoken word and sign processing in children with a CI who are exposed to signs in addition to spoken language. Study 1 assessed rapid word and sign learning in 13 children with a CI and found that performance in both language modalities correlated positively. Study 2 tested the effects of using sign-supported speech on spoken word processing in eight children with a CI, showing that simultaneously perceiving signs and spoken words does not negatively impact their spoken word recognition or learning. Together, these two studies suggest that sign exposure does not necessarily have a negative effect on speech processing in some children with a CI.

Introduction

In many countries nowadays, the majority of deaf children receive a cochlear implant (CI), and, as a result, they have greater access to spoken language. Converging evidence for the positive effect of early implantation on spoken language development and the introduction of newborn hearing screening programs have furthermore resulted in a sharp decrease in the age at which deaf children receive a CI. Implantation within the first year of life is becoming standard practice in many countries.

When the first prelingually deaf children received a CI in the 1980s, it was unknown whether they would be able to acquire spoken language from the relatively poor auditory input provided by the implant (Svirsky, McConkey Robbins, Kirk, Pisoni, & Miyamoto, 2000). In the meantime, many studies have shown that some children with a CI show similar, or even faster, rates of spoken language development compared with age-matched children with normal hearing (e.g., Geers, Nicholas, & Moog, 2007; Niparko et al., 2010; Peterson, Pisoni, & Miyamoto, 2010). However, individual outcomes are highly variable and many different factors affect the benefits a child might obtain from the CI, making it impossible to reliably predict outcomes (e.g., Belzner & Seal, 2009; Bouchard, Ouellet, & Cohen, 2008; Geers, Nicholas, & Moog, 2007; Niparko et al., 2010). Given the clearly established benefits of cochlear implantation for spoken language development, the role of sign exposure at home and at school is much debated (e.g., Delore, Robier, Bremont, Beutter, & Ployet, 1999; Geers, 2006; Knoors & Marschark, 2012; Kushalnagar et al., 2010; Leigh, 2008). At the heart of this debate is the question as to how manual communication affects spoken language development in children with a CI. A large number of studies have addressed this question by comparing children in Oral Communication (OC) settings, where only spoken language is used, to children in Total Communication (TC) settings, where both spoken and manual communication is used.
(TC) settings, where both spoken language and some form of signed communication are used. The language domains investigated in these studies comprise speech production (e.g., Tobey et al., 2007; Tobey, Geers, Brenner, Altuna, & Gabbert, 2003), speech perception (e.g., Archbold et al., 2000; Bergeson, Pisoni, & Davis, 2005; Geers, Brenner, & Davidson, 2003), vocabulary knowledge (e.g., Connor, Hieber, Arts, & Zwolan, 2000; El-Hakim et al., 2001; Kirk, Miyamoto, Ying, Perdew, & Zuganelis, 2000; Svirsky et al., 2000), reading outcomes (e.g., Connor & Zwolan, 2004), and more general expressive and receptive spoken language abilities (e.g., Geers, Nicholas, & Sedey, 2003; Kirk et al., 2000; Percy-Smith, Cayé-Thomasen, Breinegaard, & Jensen, 2010; Svirsky et al., 2000).

The findings have been somewhat contradictory, however, with many studies reporting an OC advantage, whereas others find no effect or even a TC advantage (for discussion, see e.g., Geers, 2006). Positive results have been explained by the suggestion that signed vocabulary that is acquired preimplantation might bootstrap spoken vocabulary development (Yoshinaga-Itano, 2006) and provide early language stimulation (Connor et al., 2000). In contrast, Pisoni et al. (1999) suggested that the efficiency of auditory short-term memory processes such as encoding and rehearsal might benefit from increased exposure to speech (see also Conway, Pisoni, & Kronenberger, 2009). Moreover, simultaneously attending to two visual sources of information (i.e., manual-visual and audiovisual) might create competition for limited processing resources (Bergeson et al., 2005; Burkholder & Pisoni, 2006). Finally, using sign language before implantation might stimulate cross-modal reorganization of the auditory cortex, which may negatively impact speech processing (Giraud & Lee, 2007).

Interestingly, only a few studies have compared spoken language and signed language abilities in the same children, and these have often been case studies. For instance, Klatter-Folmer et al. (2006) analyzed language samples from six deaf children of deaf and hearing parents over a period of 3 years. Three children received a CI in the course of the study. The authors concluded that the development in both language modalities was intertwined in these children (cf. Coerts et al., 1994). In fact, syntactic complexity was highest for mixed utterances, that is, utterances in which both speech and signs are used. Wiefferink et al. (2008) analyzed language samples from six children with a CI over a period of 3 years. The number of words and spoken utterances as well as the number of signs and signed utterances increased in the course of the study, but slightly faster for the spoken modality. Initially, mean length of utterance was longer in signed utterances than in spoken utterances, but by the end of the study it was the opposite (cf. Cassandro, Nicastri, Chiarella, Genovese, & Gallo, 2003). Recently, Seal, Nussbaum, Belzner, Scott and Waddy-Smith (2011) compared consonant and sign phoneme acquisition in 22 implanted children. Phonological acquisition in both language modalities followed expected developmental sequences. Importantly, cumulative consonant and sign phoneme growth were significantly correlated over time. These studies thus suggest that signing experience does not impede spoken language development.

The two studies reported here adopt a similar within-group design approach by investigating the relationship between spoken word and sign processing in a small group of children with a CI who are exposed to signs in addition to spoken language. The aim is to examine whether exposure to sign language influences their spoken word processing. To this end, Study 1 investigated the relationship between spoken word and sign learning. Study 2 investigated the effects of simultaneously perceiving signs and spoken words on spoken word recognition and learning.

It is important to emphasize that all children were exposed to signs in addition to spoken language. This is different from studies comparing children with sign exposure to children without sign exposure (e.g., children from TC and OC settings). Crucially, however, the children in the present studies varied in whether they were only exposed to sign-supported speech or also a sign language. This variation in sign exposure was expected to translate into varying signing experience for the children and as such facilitated studying the relationship between individual differences in spoken word and sign learning in Study 1. If signing experience has a direct negative effect on speech processing by children with a CI, then children that show higher sign learning performance are expected to show lower
spoken word learning performance. That is, negative correlations between sign and spoken word learning would be predicted in Study 1 (cf. Seal et al., 2011).

Word and sign learning tasks were preferred over expressive or receptive vocabulary measures, because the former are less dependent on language experience and directly assess word and sign learning abilities, whereas vocabulary measures only assess the number of words or signs already learned at a particular age (Kan & Kohnert, 2008; Prezbindowski & Lederberg, 2003; Tomblin, Barker, & Hubbs, 2007). More specifically, children in the present study were taught novel words and signs that differed in only one phonological segment, that is, minimal pairs. The ability to encode phonetic detail in novel words is a hallmark in early lexical development and has been shown to be related to vocabulary development in younger children (for discussion, see e.g., Stoe-Gammon, 2011; Swingley, 2009). Furthermore, it was expected that a novel minimal pair learning task would be more sensitive to individual differences in the speech perception abilities of children with a CI than a task that involved learning phonologically dissimilar novel words (see also Havy, Nazzi, & Bertoncini, 2013; Houston, Stewart, Moberly, Hollich, & Miyamoto, 2012).

Whereas Study 1 looked at spoken word and sign learning separately, Study 2 directly investigated whether sign-supported speech facilitates or hampers their speech processing. Although researchers have recently considered the role of the visual modality in language processing by children with a CI, most studies investigated the integration of auditory and visual speech information (e.g., Bergeson, Houston, & Miyamoto, 2010; Bergeson et al., 2005; Buckley & Tobey, 2011; Kirk et al., 2007; Most, Rothen, & Luntz, 2009; Schorr, Fox, Van Wassenhove, & Knudsen, 2005). The integration of auditory and visual input from the hands, such as signs or cospeech gestures, has to our knowledge not yet been studied in children with a CI.

Behavioral and neuroimaging studies have shown that cospeech gestures, that is, facial and hand movements that accompany speech, are tightly integrated with auditory input in language comprehension in normally hearing children and adults (e.g., Habets, Kita, Shao, Özyurek, & Hagoort, 2010; Kelly, Özyurek, & Maris, 2010; Skipper, Goldin-Meadow, Nusbaum, & Small, 2009; for a review see Kelly, Manning, & Rodak, 2008). Moreover, cospeech gestures have been found to support word learning in a foreign language in both children and adults (e.g., Kelly, McDevitt, & Esch, 2009; Tellier, 2008; but see also Kelly & Lee, 2012) and are used to support communication in children with language learning difficulties (e.g., Capone & McGregor, 2004). These studies suggest that children with a CI might benefit from the combination of signs and speech.

In that respect, there is some evidence that exposing deaf or hard-of-hearing children and adults to words and signs simultaneously might enhance spoken word processing. For instance, Hamilton and Holzman (1989) found that deaf and hearing individuals with both sign and speech experience recalled stimuli that were simultaneously spoken and signed better than stimuli that were only spoken or only signed. In addition, Emmorey, Petrich and Gollan (2012) recently showed that hearing signers are faster to make semantic decisions when presented with simultaneous productions of speech and sign in comparison to words or signs alone. Language production studies furthermore suggest that hearing signers integrate both modalities effortlessly and without confusion (Emmorey, Borinstein, Thompson, & Gollan, 2008; Kovelman et al., 2009). Most relevant to the present studies, Mollink, Hermans and Knoors (2008) examined the effects of using signs in spoken vocabulary training for children with a mild-to-moderate hearing loss and found that signs had a positive effect on the learning and retention of new spoken vocabulary. Specifically, pictures that were trained with words and signs combined received the highest percentage correct scores.

Alternatively, perceiving signs and spoken words simultaneously may interfere with spoken word processing. Bergeson et al. (2005) found that in the early test intervals in their study children in OC settings outperformed children in TC settings. They suggested that the latter might have to distribute their attention over two visual sources of information (i.e., manual-visual and audiovisual). Such division of attention could create competition between limited processing resources in working memory and result in less efficient speech processing (see also Burkholder & Pisoni, 2006). However, this possibility has not yet been empirically tested.
Study 1

Participants

Thirteen prelingually deaf 5- to 6-year-old children with a CI (3 girls, 10 boys) participated in this study, after consent was obtained from their parents and teachers. Two other children were tested but later excluded. Individual background information for the 13 children is provided in Table 1. Their mean age was 5 years 9 months (4 years 4 months to 6 years 7 months, SD = 10 months). None of the children in the sample were known to have additional disabilities. For all children the surgery was uneventful with full insertion of the implants and they were fitted with the latest speech-processing algorithm available at the time. All children wore their implant for the greater part of the day. Parent involvement was overall average to high. All children had Dutch as their native language. The majority of these children (10) had previously participated in a longitudinal study that investigated the development of auditory perception, speech intelligibility, and receptive and expressive spoken language abilities of 18 children with a CI from the Netherlands and Flanders (the Dutch-speaking part of Belgium) from shortly before implantation until 3 years post implantation, approximately one year before the present study (Wiefferink et al., 2008; Wiefferink, 2012). Concurrent measures of auditory and spoken language abilities of the children were unfortunately not available to the authors.

All children had received their implant before their fourth birthday, and the mean age at implantation in the sample was 1 year 9 months (0 years 7 months to 3 years 9 months, SD = 12 months). Five of the 13 children had received their implants before 12 months of age and eight of the 13 children had received their implants before 24 months of age. On average, they had been using their CI for 4 years (1 year 7 months to 5 years 11 months; SD = 13 months).

The background of the children from the Netherlands and Flanders differed in several important respects. Firstly, because of earlier introduction of newborn hearing screening in Flanders the mean age at implantation for the latter was 2 years 3 months, whereas for the children from the Netherlands it was 1 year 2 months. Secondly, bilateral implantation as well as the use of CIs in combination with acoustic hearing aids is more common in Flanders than in the Netherlands. At the time of study, three children from Flanders had received a second CI and one had been fitted with an acoustic hearing aid for the nonimplanted ear. One child from the Netherlands had received a second CI, but wore it infrequently.

Furthermore, the children from the Netherlands and Flanders also differed in the amount and nature of sign exposure, due to different perspectives on the role of sign language in education between the countries. Both groups of children were taught through sign-supported speech, that is, simultaneous use of spoken language accompanied by signs from the surrounding

Table 1 Background characteristics of the children with a cochlear implant in Study 1

<table>
<thead>
<tr>
<th>Participant</th>
<th>Gender</th>
<th>Age at implantation</th>
<th>Stimulation</th>
<th>Implant type</th>
<th>Educational setting</th>
</tr>
</thead>
<tbody>
<tr>
<td>N7</td>
<td>F</td>
<td>5,1</td>
<td>0,7</td>
<td>CI</td>
<td>SSD+NGT</td>
</tr>
<tr>
<td>X5</td>
<td>M</td>
<td>6,7</td>
<td>0,7</td>
<td>CI+HA</td>
<td>Cochlear (Sprint)</td>
</tr>
<tr>
<td>A1</td>
<td>M</td>
<td>5,3</td>
<td>0,9</td>
<td>CI</td>
<td>Cochlear (Sprint)</td>
</tr>
<tr>
<td>J3</td>
<td>M</td>
<td>4,4</td>
<td>0,10</td>
<td>CI</td>
<td>Cochlear (Sprint)</td>
</tr>
<tr>
<td>V4</td>
<td>M</td>
<td>6,7</td>
<td>0,11</td>
<td>CI</td>
<td>Cochlear (Freedom)</td>
</tr>
<tr>
<td>S7</td>
<td>M</td>
<td>5,2</td>
<td>1,2</td>
<td>BI</td>
<td>Cochlear (Sprint)</td>
</tr>
<tr>
<td>T1</td>
<td>M</td>
<td>4,10</td>
<td>1,11</td>
<td>CI</td>
<td>Cochlear (Sprint)</td>
</tr>
<tr>
<td>L2</td>
<td>F</td>
<td>6,0</td>
<td>2,0</td>
<td>CI</td>
<td>Clarion (Platinum)</td>
</tr>
<tr>
<td>D8</td>
<td>M</td>
<td>6,7</td>
<td>2,1</td>
<td>BI</td>
<td>Cochlear (Freedom)</td>
</tr>
<tr>
<td>K3</td>
<td>M</td>
<td>6,4</td>
<td>2,1</td>
<td>CI</td>
<td>Clarion (Platinum)</td>
</tr>
<tr>
<td>L6</td>
<td>F</td>
<td>6,7</td>
<td>2,9</td>
<td>BI</td>
<td>Digisonic (SP)</td>
</tr>
<tr>
<td>L4</td>
<td>M</td>
<td>6,7</td>
<td>3,2</td>
<td>CI</td>
<td>Cochlear (Sprint)</td>
</tr>
<tr>
<td>S5</td>
<td>M</td>
<td>5,4</td>
<td>3,9</td>
<td>CI</td>
<td>Cochlear (Freedom)</td>
</tr>
</tbody>
</table>

Note. Ages are in years, months; CI, cochlear implants; SSD, sign-supported Dutch; NGT = Nederlandse Gebarentaal (Sign language of the Netherlands); HA = hearing aid.
sign language (also referred to as Simultaneous Communication). In addition, the children from the Netherlands were taught Sign Language of the Netherlands (NGT, Nederlandse Gebarentaal) as a subject in school. Furthermore, their parents had followed courses on NGT (on average about 30 lessons), and exposure to NGT was provided in daycare and preschool settings. Two children in this sample (one from Flanders, one from the Netherlands) already attended mainstream education at the time of this study. These children were no longer exposed to signs at school.

Importantly, this situation is different from studies comparing children in OC and TC settings. Firstly, it is typically assumed that the children in an OC setting are not exposed to signs at all. Secondly, the label TC covers a great variety of practices including the use of sign language and sign-supported speech and, for instance, also sign systems such as Seeing Exact English (Spencer & Tomblin, 2006). Importantly, the more gradient variation in the amount of sign exposure in the present study allowed us to look at the relationship between individual differences in spoken word and sign processing for the same children.

Nonword Picture-Matching

**Stimuli.** Twelve monosyllabic consonant-vowel-consonant nonwords were created using WordGen© (Duyck, Desmet, Verbeke, & Brysbaert, 2004), a (non)word generator program based on the CELEX database (Baayen, Piepenbrock, & Van Rijn, 1993). The minimal nonword pairs were formed contrasting either in the vowel (/ɑ/-/a/ or /ɪ/-/i/) or initial consonant (/f/-/s/ or /b/-/p/), resulting in three pairs per sound contrast (see Appendix A). In addition, four monosyllabic familiar words known to typically developing 6-year-old children were selected as control stimuli (Schaerlaekens, Kohnstamm, & Lejaegere, 1999). All stimuli were embedded in short carrier phrases (see below) and recorded in a sound-attenuated room with a sampling rate of 44.1kHz and converted to the WAVE (32-bit linear pulse code modulation) format. The pictures used in the task were black-and-white drawings of novel and familiar objects: The pictures of novel objects were selected from a previously used database (Escudero, Broersma, & Simon, 2012; Escudero, Hayes-Harb, & Mitterer, 2008; Shatzman & McQueen, 2006), and the pictures of familiar objects were taken from a publicly available picture set designed for reading instruction in classrooms.

**Task.** The nonword picture-matching task used in this study was based on rapid word learning designs in which children have to learn novel words after only a few exposures to the word and referent (see e.g., Lederberg, Spencer, & Prezbindowski, 2000 for discussion). Such tasks have been previously used in studies with young children with a CI, although these tested sets of phonologically dissimilar nonwords (e.g., Tomblin et al., 2007; Willstedt-Svensson, Löfqvist, Almqvist, & Sahlén, 2004). A similar, but extended design to the one used here has been used by Escudero et al. (2012) to assess adult second language learners’ ability to learn novel minimal pairs.

The task consisted of four blocks, corresponding to four stimulus sets of two novel words/objects and one familiar word/object. Each block consisted of a familiarization part and a testing part. Presentation of the four blocks was counterbalanced across children and separated by a brief pause. To control for potential effects of iconicity between the words and the pictures as well as for stimulus-specific nonword preferences, the three different nonwords pairs created for each contrast in the task were alternated between children.

During familiarization, each word/object pair was presented three times in a random order in the carrier phrase ‘Kijk, een X!’ (Look, a X!). During testing, one of the familiarized (non)words was first presented in the carrier phrase ‘Waar is de X?’ (Where is the X?), followed by two of the pictures on the left and right side of the screen, which remained visible until a left or right response key (indicated by stickers) was pressed. The children responded to four target (two novel objects) and four control (one familiar object, one novel object) trials in random order. In the four target trials, each novel object was tested twice. In the four control trials, the novel and familiar objects were each tested twice. Presentation on the screen (left or right side) was counterbalanced for both novel and familiar objects.

E-Prime 2.0® (Psychology Software Tools, Pittsburgh, PA) was used to present the stimuli and record responses and reaction times. Reaction times
were measured from the offset of the auditory stimulus to the overt response, that is, the key press. A practice block with two phonologically dissimilar nonwords and a familiar word preceded the experiment. The familiarization part was identical to the other blocks, but the testing part was limited to three trials, two target trials, and one control trial presented in random order. The task took approximately 15 min.

Nonsign Picture-Matching

Stimuli. Six minimal nonsign pairs were formed contrasting either in hand configuration (/open/-/closed/) or location (/eye/-/chin/), three pairs for each contrast (see Appendix B). All nonsigns were checked by native NGT and VGT (Vlaamse Gebarentaal, Flemish Sign Language) signers to ensure they were indeed possible but nonexisting signs. Two familiar signs that are cognate signs in NGT and VGT were selected as control stimuli from NGT teaching material for young deaf children because ratings of signs for age of acquisition are not available for NGT or VGT. All stimuli were embedded in short carrier phrases (see below) and recorded against a blue-grey background. Pinnacle® Studio 11 was used to digitally capture and compress the recorded clips to Windows Media Video (WMV) format (440 kbps, 25 fps, 360 × 280 pixels). The pictures used in the task were selected from the same databases as for the nonword picture-matching task.

Task. The design of the nonsign picture-matching task was similar to that of the nonword picture-matching task. The experiment was divided into two blocks, corresponding to two stimulus sets of one minimal nonsign pair and one familiar sign. Presentation of the two blocks was counterbalanced across children and separated by a brief pause. As in the nonword picture-matching task, the three different nonsign pairs created for each contrast in the task were alternated between children.

The monosyllabic nonsigns and familiar signs were presented in the carrier phrase ‘kijk, x!’ (see, x!) during familiarization, and ‘where x?’ (where x?) during testing. In all other respects, familiarization and testing was as in the nonword picture-matching task, that is, nine familiarization trials followed by eight two-alternative forced-choice identification trials. In the familiarization trials and the testing trials, the novel objects were presented 25% upwards from the center of the screen and the video stimuli 25% downwards.

E-Prime 2.0® (Psychology Software Tools, Pittsburgh, PA) was used to present the stimuli and record responses and reaction times. Similar to the nonword picture-matching task, a practice block with two phonologically dissimilar nonsigns and a familiar sign preceded the experiment. The task took approximately 10 min.

Procedure

Testing took place individually in a quiet room at the children’s school. Children performed both tasks on the same day, with the nonword picture-matching task always administered first. Instructions were in spoken language supported with signs for all children. The tasks were presented on a DELL® Latitude D630 laptop using two external speakers (Trust® SP-2310). The nonword picture-matching task was presented at a sound level within each child’s own range of comfort determined during the practice.

Results

Figures 1 and 2 show the mean overall percentage correct scores and reaction times, respectively, in both language modalities and for both target and control trials. A 2 (Modality) × 2 (Trial type) repeated measures analysis of variance (ANOVA) on accuracy scores with Modality and Trial Type as within-subjects variables revealed a main effect of Trial type (F[1,12] = 50.54, p < .01). Unexpectedly, scores were higher for the relatively easy control trials (one novel and one familiar object) than the more difficult target trials (two novel objects) in both language modalities. Although scores for target trials appeared to be higher for the signed (M = 64.4% correct) than the spoken modality (M = 52.2% correct), the Modality × Trial type interaction did not approach significance (F[1,12] = 1.48, p = .25). Similar to the accuracy analysis, a 2 (Modality) × 2 (Trial type) repeated measures ANOVA on reaction times only revealed a main effect of Trial type (F[1,12] = 6.47, p < .05). Responses were faster on control than target trials in both language modalities.
Figure 1  Mean % correct scores on target and control trials in the nonword (speech) and nonsign (sign) picture-matching task. Error bars represent one standard error from the mean.

Figure 2  Mean reaction times (ms) on target and control trials in the nonword (speech) and nonsign (sign) picture-matching task. Error bars represent one standard error from the mean.
The error bars in the figures show more individual variation for sign versus spoken word learning. This was expected given the variation in sign exposure for the children. To examine whether individual differences in sign and spoken word learning performance were positively or negatively related to each other, a correlation analysis was performed between scores and reaction times in both modalities. Both scores and reaction times in the nonword and nonsign picture-matching tasks correlated positively ($r = .67$, $p < .05$ and $r = .92$, $p < .01$, respectively), whereas negative correlations would have been expected if signing experience had negatively impacted spoken word learning for the children in our sample. Instead, these results suggest a positive relationship between the two language modalities for the small group of children in this study (see also Seal et al., 2011). It is important to note that correlations do not imply causality. The present results, therefore, should not be interpreted as evidence that good sign learning causes good spoken word learning, or vice versa. They only suggest that the children with higher sign learning performance were the same children that showed higher spoken word learning performance. This point will be further addressed in the Discussion.

Study 2

Instead of examining spoken word and sign processing separately, Study 2 investigated spoken word processing in the context of sign-supported speech. Many children with a CI, including those in the present studies, are exposed to this form of sign support, and sign-supported speech forms a core component of many Total Communication programs (e.g., Spencer & Tomblin, 2006). Importantly, it has been suggested that perceiving sign-supported speech may interfere with speech processing by children with a CI (e.g., Bergeson et al., 2005). To examine this possibility, a new multimodal picture-matching task was designed based on the tasks used in Study 1. Novel and familiar word and sign stimuli were included to assess effects on spoken word learning and recognition, respectively. Furthermore, the role of phonological processing load was addressed by including both minimal and nonminimal word pairs as stimuli.

Participants

Approximately sixteen months after Study 1, the parents of the children that had participated in Study 1 were asked by letter for their consent to include their child in a follow-up study. Consent was obtained from the parents of eight children. Individual background information for these children is provided in Table 2. Their mean age at testing was 6 years 11 months and their mean age at implantation was 1 years 10 months. At the time of testing, three of the children attended mainstream education and five children attended schools for the deaf.

Stimuli

The stimuli in the experiment consisted of video recordings of familiar and novel words, signs, and code-blends, that is, simultaneous productions of spoken words and signs (Emmorey et al., 2008). All stimuli were embedded in short carrier phrases and recorded against a blue background to optimize sign visibility using an external microphone attached to the video.

Table 2  Background characteristics of the children with a cochlear implant in Study 2

<table>
<thead>
<tr>
<th>Participant</th>
<th>Gender</th>
<th>Age</th>
<th>Age at implantation</th>
<th>Stimulation</th>
<th>Implant type</th>
<th>Educational setting</th>
</tr>
</thead>
<tbody>
<tr>
<td>N7</td>
<td>F</td>
<td>6,6</td>
<td>0,7</td>
<td>CI</td>
<td>Clarion (Platinum)</td>
<td>Mainstream</td>
</tr>
<tr>
<td>A1</td>
<td>M</td>
<td>6,8</td>
<td>0,9</td>
<td>CI</td>
<td>Cochlear (Sprint)</td>
<td>Mainstream</td>
</tr>
<tr>
<td>J3</td>
<td>M</td>
<td>5,9</td>
<td>0,10</td>
<td>CI</td>
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<td>SSD</td>
</tr>
<tr>
<td>S7</td>
<td>M</td>
<td>6,7</td>
<td>1,2</td>
<td>Bilateral CI</td>
<td>Cochlear (Sprint)/Digisonic (SP)</td>
<td>SSD</td>
</tr>
<tr>
<td>L2</td>
<td>F</td>
<td>7,4</td>
<td>2,0</td>
<td>CI</td>
<td>Clarion (Platinum)</td>
<td>SSD+NGT</td>
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<td>D8</td>
<td>M</td>
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<td>2,1</td>
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<td>Cochlear (Sprint/Freedom)</td>
<td>Mainstream</td>
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<td>M</td>
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<td>CI</td>
<td>Cochlear (Freedom)</td>
<td>SSD+NGT</td>
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Note. Ages are in years, months; CI, cochlear implant; SSD, sign-supported Dutch; NGT = Nederlandse Gebarentaal (sign language of the Netherlands).
camera, and digitally captured, and compressed to WMV format (440 kbps, 25 fps, 360 × 280 pixels) using Pinnacle® Studio 11.

Four nonword pairs and four familiar word pairs were selected, half of which were minimal pairs and half of which were nonminimal pairs. The two nonminimal nonword pairs were created by randomly pairing phonologically different nonwords that had already been used in Study 1 (see Appendix A). Two new minimal nonword pairs were formed that contrasted in the words medial vowel (/tuk/-/tik/ and /fup/-/fip/). The vowel contrast /u/-/i/ was chosen because it has a strong visual correlate and thus allowed us to test the prediction that manual-visual and auditory-visual input compete for processing resources (Bergeson et al., 2005). Similar to the other nonwords, they conformed to a monosyllabic consonant-vowel-consonant structure. Furthermore, two familiar minimal word pairs (/kɔp/-/pɔp/ “mug-doll” and /tuk/-/zɔk/ “branch-sack”) and two familiar nonminimal word pairs (/muw/-/dos/ “sleeve-box” and /touw/-/ros/ “rope-rose”) were selected that should be known to typically developing 6-year-old children (Schaerlaeken et al., 1999).

Two minimal nonsign pairs that contrasted in hand configuration (/open/-/closed/) were selected from the set of nonsigns that had already been used in Study 1 (see Appendix B). Additionally, two nonminimal nonsign pairs were formed (see Appendix B). Two familiar nonminimal sign pairs that were cognate signs for NGT and VGT were selected from teaching materials for deaf children (beer “bear,” boek “book,” bril “glasses,” pet “cap”). Familiar minimal sign pairs were not included in the study.

In order to create the familiar code-blend stimuli, the above-mentioned familiar minimal and nonminimal word pairs were also recorded as code-blends, that is, simultaneously produced with the corresponding sign translations. As a consequence, in the familiar code-blend pairs constructed from familiar minimal word pairs, only the words but not the signs were minimally different. This is because it is very difficult to find a familiar minimal word pair for which the sign translations also form a minimal pair. To create the novel code-blend stimuli (“noncode-blends”), each minimal nonsign pair was randomly paired with one of the minimal nonword pairs, and each nonminimal nonsign pair was randomly paired with one of the nonminimal nonword pairs and recorded as code-blends. That is, different from the familiar code-blend pairs, for the novel code-blend pairs both the words and the signs formed minimal pairs or nonminimal pairs.

The picture stimuli were black-and-white drawings of novel and familiar objects from the same picture databases as used in Study 1.

Multimodal Picture-Matching Task

The task consisted of six blocks, distributed across three conditions: speech, sign, and sign-supported speech. Each condition included a block with a relatively low phonological processing load (nonminimal novel and familiar pairs) and a block with a relatively high phonological processing load (minimal novel and familiar pairs). Similar to Study 1, each block consisted of a familiarization and a testing part.

During familiarization, the children were presented three times with two novel and two familiar objects in speech, sign, or both. The carrier phrases were “Kijk, een X!” (Look, a X!) for the speech condition, “INDEX X!” for the sign condition, and both for the sign-supported speech condition. In each trial, a picture and a movie were presented side by side in the center of the screen, with the picture always presented on the left.

Following familiarization, a black-and-white blocked flag was displayed in the center of the screen for 2,000 ms in order to fixate attention to the center. Next, 12 two-alternative forced-choice identification trials were presented in random order. During testing, one of the familiarized (non)words or (non)signs was presented in the carrier phrase “Waar is de X?” (Where is the X?) and “WAAR X?” (WHERE X?), respectively. The movie stimulus was presented in the center of the screen, followed by two pictures, one at the left and one at the right side of the screen, which remained visible until a left or right response key (indicated by stickers) was pressed. The children responded to four novel (two novel objects), four familiar (two familiar objects), and four control (one novel object, one familiar object) trials during testing. In total, each object was tested three times. Side of presentation (left or right side) of the pictures on the screen was counterbalanced.
Importantly, during testing in the sign-supported speech condition, children were first tested on the spoken words only. They were separately tested on the signs after completing all spoken word testing trials. If recognition and learning of code-blends had been tested, it would have been difficult, if not impossible, to establish whether the children primarily responded to the words or the signs in the code-blends during testing. The disadvantage of testing the spoken words before the signs is that the familiarization-testing interval is longer for the latter. As a result, performance on the signs in the sign-supported speech condition might be poorer than performance on the signs in the sign condition simply because more time had elapsed between familiarization and testing. However, the advantage of this design is that the testing phase for the words in the sign-supported speech condition was identical to that in the speech condition. That is, the only difference between the two conditions was whether sign-supported speech was used during familiarization.

E-Prime 2.0® (Psychology Software Tools, Pittsburgh, PA) was used to present the stimuli and record accuracy and reaction times. Reaction times were measured from the offset of the video stimulus to the overt response, that is, the key press. A practice block in speech with two phonologically dissimilar nonwords and two phonologically dissimilar familiar words preceded the experiment. Familiarization was identical to the other blocks, but testing was limited to six trials, two for each type of testing trials, presented in random order. The task took approximately 25 min.

Procedure

The experiment consisted of six blocks, with three conditions (speech, sign, and sign-supported speech). In the first block of each condition, the relatively easy novel and familiar nonminimal pairs were presented, followed by a block with the more difficult novel and familiar minimal pairs. Before the start of a new block, children were reminded of the condition that would follow next. Before completing the sign-supported speech condition, they were made aware that they would be tested on the words and signs separately. Half of the children completed the speech before the sign condition, and half completed the sign before the speech condition. All children completed the sign-supported speech condition last. This was done to ensure recent exposure to both language modalities before completing this condition. The order of the speech and sign condition was counterbalanced to account for potential priming effects in the sign-supported speech condition from the preceding condition.

Furthermore, presentation of word and sign pairs in the three conditions was counterbalanced across children to the extent possible. That is, half of the children were presented with word pairs A and B in the speech condition and with the word pairs C and D in the sign-supported speech condition, and half with pairs C and D in the speech condition and pairs A and B in the sign-supported speech condition. Because it was impossible to counterbalance both the familiar word and sign pairs this way (this would have resulted in presentation of the same pictures in the speech and sign conditions), presentation of the familiar sign pairs was not counterbalanced.

Results

Table 3 provides the descriptive statistics of the percentage correct scores and reaction times in all conditions (speech, sign, and sign-supported speech, the latter separated for spoken words and signs). Figures 3 and 4 show the mean percentage correct scores and reaction times for spoken word recognition in the speech and sign-supported speech conditions. The bars are clustered according to stimulus type (familiar or novel and phonologically similar or different). Scores and reaction times for spoken words in the speech and sign-supported speech conditions, and for signs in the sign and sign-supported speech conditions, were statistically compared using nonparametric related samples Wilcoxon Signed Rank tests to accommodate for the small sample size.

None of the planned related-sample comparisons between scores for spoken words in the speech and sign-supported speech conditions or between scores for signs in the sign and sign-supported speech conditions approached significance (all $p > .15$).

Planned related-sample comparisons between reaction times for spoken words in the speech and sign-supported speech conditions showed a trend for
faster responses to familiar minimal word pairs in the sign-supported speech condition than the speech condition (sign-supported speech: median = 1,010 ms, speech: median = 1,630 ms, Z = 1.960, p = .05). None of the other comparisons approached significance (all p > .09). Planned related-sample comparisons between reaction times for signs in the sign and sign-supported speech conditions further showed that the children responded significantly slower to familiar sign pairs in the sign-supported speech condition than the sign condition in the block that also included minimal novel sign pairs (sign-supported speech: median = 1,614 ms, sign: median = 1,027 ms, Z = 2.240, p < .05). None of the other comparisons approached significance (all p > .09).

In sum, sign-supported speech did not have a negative impact on spoken word processing in the small sample that participated in the two studies. Simultaneously perceiving signs and spoken words did not interfere with the learning of novel words or retrieval of familiar

Table 3  Means and standard deviations (between parentheses) of percentage correct scores (%) and reaction times (RT) in Study 2

<table>
<thead>
<tr>
<th>Stimulus type</th>
<th>Trial type</th>
<th>Speech</th>
<th>Sign</th>
<th>Sign-supported speech</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td>Speech</td>
<td>Sign</td>
<td>Speech</td>
</tr>
<tr>
<td>Minimal pairs</td>
<td>Familiar</td>
<td>% 90.6 (18.6)</td>
<td>96.9 (8.8)</td>
<td>96.9 (8.8)</td>
</tr>
<tr>
<td></td>
<td></td>
<td>RT 1,409 (571)</td>
<td>1,173 (365)</td>
<td>1,142 (377)</td>
</tr>
<tr>
<td></td>
<td>Novel</td>
<td>% 84.4 (12.9)</td>
<td>78.1 (20.9)</td>
<td>93.8 (11.6)</td>
</tr>
<tr>
<td></td>
<td></td>
<td>RT 1,585 (501)</td>
<td>2,235 (910)</td>
<td>1,566 (475)</td>
</tr>
<tr>
<td></td>
<td>Control</td>
<td>% 96.9 (8.8)</td>
<td>96.9 (8.8)</td>
<td>93.8 (11.6)</td>
</tr>
<tr>
<td></td>
<td></td>
<td>RT 1,524 (502)</td>
<td>1,289 (663)</td>
<td>1,305 (403)</td>
</tr>
<tr>
<td>Nonminimal pairs</td>
<td>Familiar</td>
<td>% 100.0 (0.0)</td>
<td>93.8 (11.6)</td>
<td>100.0 (0.0)</td>
</tr>
<tr>
<td></td>
<td></td>
<td>RT 1,551 (935)</td>
<td>1,165 (236)</td>
<td>1,018 (333)</td>
</tr>
<tr>
<td></td>
<td>Novel</td>
<td>% 78.1 (24.8)</td>
<td>93.8 (11.6)</td>
<td>62.5 (37.8)</td>
</tr>
<tr>
<td></td>
<td></td>
<td>RT 2,101 (1126)</td>
<td>1,453 (501)</td>
<td>2,086 (1196)</td>
</tr>
<tr>
<td></td>
<td>Control</td>
<td>% 93.8 (17.7)</td>
<td>100.0 (0.0)</td>
<td>96.9 (8.8)</td>
</tr>
<tr>
<td></td>
<td></td>
<td>RT 1,356 (363)</td>
<td>1,335 (394)</td>
<td>1,425 (352)</td>
</tr>
</tbody>
</table>
words among this group of children. Furthermore, this was true for phonologically similar as well as phonologically dissimilar word pairs.

Because the sample size in Study 2 was very small, the results should be interpreted with caution, especially because these concern null effects. However, a visual inspection of the data shows that, except for novel nonminimal word pairs, scores for the two conditions were either similar or numerically higher for the sign-supported speech condition. Similarly, reaction times for the two conditions were either similar or numerically lower for the sign-supported speech condition. Indeed, the individual data showed that six out of eight children showed faster reaction times for words in the sign-supported speech condition compared to the speech condition for familiar minimal and nonminimal pairs, and five out of seven for novel nonminimal pairs (one child had switched the labels of the words and therefore no correct trials could be analyzed for reaction times). Similarly, only one out of eight children scored lower on words in the sign-supported speech than speech condition for familiar minimal word pairs, zero out of eight for familiar nonminimal word pairs, and two out of eight for novel minimal word pairs. The rest of the children improved or obtained equal scores in the two conditions. The only word pairs which proved particularly challenging for the children were novel nonminimal word pairs, for which four out of eight children scored lower and responded slower on the sign-supported speech condition compared to the speech condition. The individual patterns of the children in this study suggest that sign-supported speech did not negatively affect their speech processing. Further evidence is needed using a sample size with sufficient power and including a control group to reveal the unique relationship between sign-supported speech and lexical processing in children using CIs.

Discussion

The two studies reported here provide new insight into the relationship between speech and sign processing for children with a CI who are exposed to signs in addition to spoken language. Study 1 demonstrated that sign learning may not have had the direct negative impact on the spoken word processing of the children studied that had been predicted by previous research. Study 2 suggests that, at least for the children in this study, sign-supported speech did not interfere with spoken word processing and may even have provided a
benefit to the children as they were trying to perceive perceptually confusable words.

The children in Study 1 did not experience the negative effect of sign exposure on their spoken language and speech processing as predicted by previous research (e.g., Bergeson et al., 2005; Burkholder & Pisoni, 2006; El-Hakim et al., 2001; Geers, et al., 2003; Kirk et al., 2000; Pisoni et al., 1999). However, several important differences between studies could explain the divergent findings. Firstly, and most notably, the present study investigated both sign and spoken word learning in the same children, whereas other studies investigated only spoken vocabulary knowledge and compared children with and without sign exposure (e.g., children in TC settings versus OC settings). The disadvantage of a between-subjects approach is that any observed differences in spoken language outcomes between the two groups may be caused by uncontrolled differences between the two groups other than their respective educational modalities, such as nonverbal IQ, age at implantation, preimplant hearing thresholds or programming characteristics. Also, it is often unclear why parents decided for one or the other educational modality for their child. It is not unlikely, for instance, that precisely those children that are showing less than expected progress with the CI early on are more likely to later be enrolled in Total Communication settings.

An illustration of the potential impact of uncontrolled factors in studies comparing different groups of children with a CI comes from the longitudinal study on spoken language development in 18 children with a CI from the Netherlands and Flanders that was referred to in the introduction and that included 10 children that also participated in the present study (Wiefferink et al., 2008; Wiefferink, 2012). In that study, the authors compared auditory perception, speech intelligibility and expressive and receptive spoken language outcomes between the children from the Netherlands and Flanders to examine the influence of linguistic environment, more specifically the presence of sign language in the input to the children. In stark contrast to the findings presented here, the authors concluded that spoken language development progressed more rapidly in the children without a sign language in their input. However, as also acknowledged by the authors, there were several alternative explanations possible for the observed group difference. Most notably, the children from Flanders had better residual aided hearing and had received professional care at an earlier age. In fact, residual aided hearing before implantation correlated significantly with auditory perception, speech intelligibility, and both receptive and expressive spoken language ability after implantation.

The current within-subject approach is evidently safeguarded against such potential between-group differences. However, this approach has its own disadvantages, most notably the potential role of uncontrolled individual differences in cognitive skills, such as nonverbal IQ, verbal working memory, and sustained attention, which likely influence both speech and sign processing abilities for children with a CI.

A second important difference from previous studies is that all the children in our sample were exposed to signs, either in the form of sign-supported speech only or also in the form of a sign language, whereas in studies comparing children in OC and TC settings, it is usually assumed that the former receive no sign exposure at all. Although this situation allowed us to look at the relationship between individual differences in spoken word and sign processing for the same children, it makes a direct comparison between studies difficult.

Moreover, although studies comparing children in OC and TC settings have included a range of spoken language outcome measures, most studies looked at speech production and/or expressive vocabulary and language. In contrast, the present studies looked at receptive language processing, more specifically spoken word learning and recognition. Moreover, experimental tasks were used instead of standardized speech perception or receptive vocabulary tests. We did not include such assessments because these are highly dependent on previous language experience. For instance, studies with child and adult bilinguals have consistently shown smaller vocabularies in each of their languages than monolingual children and adults (Bialystok, 2009). Smaller expressive and receptive vocabularies in children with a CI who are exposed to signs in addition to spoken language are therefore not unexpected. When spoken and signed vocabulary are considered together, these children may have equal or in fact larger vocabularies than those who are only exposed to spoken language (e.g., Connor et al., 2000). Nevertheless,
this should be considered an important limitation of the present study, especially because concurrent measures of, for example, postimplant auditory perception, speech intelligibility, and receptive or expressive grammar were also not available. Future studies should certainly include a battery of standardized assessments to investigate whether the current findings generalize across children with different spoken language proficiencies and across different spoken language measures.

In addition to differences in design between the present and previous studies, the small sample size and relatively wide range in age at implantation in the present studies further limits generalizability of the results. It is therefore important that future studies using a similar within-subject approach with large samples of children with early implantation corroborate the present findings before strong conclusions can be drawn. In that respect it is important to note that Seal et al. (2011) also observed positive correlations between English consonant and ASL sign phoneme acquisition in 22 children implanted between 13 months and 84 months. In addition, Woll, Rinaldi, Woolfe, Herman, and Roy (2009) reported positive correlations between spoken and signed expressive and receptive vocabulary knowledge in 20 British bilingual deaf children. Furthermore, several studies have reported positive correlations between signing skills and reading proficiency for deaf children (e.g., Hermans, Ormel, & Knoors, 2010; Hermans, Ormel, Knoors, & Verhoeven, 2008; see also Mayberry, del Giudice, & Lieberman, 2011). However, these latter studies generally included both children with and without a CI.

The results of the two studies presented here suggest that sign-supported speech did not negatively affect spoken word processing in the small sample of children with CIs that we studied. More specifically, perceiving speech and sign at the same time does not appear to create competition between limited processing resources, as suggested by Bergeson et al. (2005). In fact, under some circumstances, namely when the auditory information that needs to be processed is particularly challenging, signs may even be beneficial, for instance, in retrieving lexical representations of phonologically similar words. These results are in line with those by Mollink et al. (2008), who also observed positive effects when adding sign to speech in spoken vocabulary training for children with a mild-to-moderate hearing loss. Importantly, the lack of significant differences between scores in the sign-supported speech and sign conditions rules out the possibility that negative effects on speech processing were not observed because the children did not look at the signs in the sign-supported speech condition.

Although not statistically significant, we would still like to suggest a possible explanation for the trend toward faster responses in the sign-supported speech condition than the speech condition for familiar phonologically similar words. It is possible that the children coactivated spoken and signed lexical representations during familiarization in the sign-supported speech condition, which might have resulted in increased lexical or semantic activation and subsequent faster retrieval of the spoken lexical representations during testing. In line with this explanation, Emmorey et al. (2012) recently showed that adult hearing signers made faster semantic decisions when the stimuli were presented as code-blends than as signs or spoken words alone. The authors suggest that coactivation of spoken and signed representations at the phonological and/or semantic level may increase lexical activation. It is furthermore possible that phonologically similar words benefit more from increased lexical and/or semantic activation than phonologically dissimilar words because the former compete more with each other during spoken word recognition (e.g., Magnuson, Dixon, Tanenhaus, & Aslin, 2007).

Evidently, further studies with larger samples are needed to examine the interaction between the spoken and signed modality during language processing, preferably over time. It is not unlikely, for instance, that any benefits from sign-supported speech are especially pronounced in the first few years following implantation and become smaller over time when children gain more experience with the CI and become more proficient in the spoken modality. Alternatively, increasing experience with perceiving (and producing) speech and sign simultaneously might lead to stronger cross-language facilitation. Future studies should also investigate whether the results observed in the present study with the presentation of isolated words and signs extend to the processing of sign-supported speech at the sentence and discourse level.
Recently, Knoors and Marschark (2012) argued in favor of a more differentiated approach to language planning and policy for deaf children in light of the increased potential to learn spoken language through early cochlear implantation. Among other things, they argued for the potential benefits of sign-supported speech, particularly for deaf children with early implantation and relatively strong spoken language proficiency. The results presented lend credence to their position. However, it is important to emphasize that sign exposure was defined rather broadly in the present study, including both sign-supported speech and a sign language. Our results do not provide evidence in favor of or against different types of sign exposure but do argue against claims in the literature and beliefs among professionals and parents that any form of sign exposure will negatively affect the spoken language abilities of CI children. The ultimate goal of cochlear implantation is arguably to provide opportunities for the acquisition and use of spoken language. Auditory stimulation and spoken language development through the implant must therefore be encouraged and optimized. However, this does not mean there is no role at all anymore for sign language or other forms of sign support. Early and continued access to language in both the visual and auditory modality is crucial to minimize the risk for language and cognitive delays (e.g., Kushalnagar et al., 2010). Our findings suggest that sign exposure was of benefit to the small group of children with CIs who participated in these studies. Additional research on a sufficiently large sample to establish at least moderate power is, however, warranted. Clearly, therefore, more studies are needed that investigate the possible benefits of sign exposure for the growing population of deaf children with a CI.

Notes

1. One child was implanted at 1 year 8 months, and fitting and programming of the device had been problematic due to behavioral difficulties. In general, she was considered a low performer with the implant and relied to a large extent on sign language in daily communication. The other child was implanted early (0 years 8 months), and fitting and programming of the implant had been unproblematic; however, this child was very inattentive during testing.

2. Averaged across consonant and vowel contrasts, scores on target trials in the nonword picture-matching task did not exceed chance level for the children with a CI. However, this mainly resulted from relatively poor performance on the consonant contrasts (Giezen, 2011).

3. Age at implantation and length of CI use were included in correlation analyses in Study 1 and Study 2. Although in both studies significant positive correlations between length of CI use and several outcome measures were observed, one child with limited hearing experience (less than 2 years) mainly drove these correlations, and they are for that reason not reported. Age at implantation did not correlate significantly with any of the outcome measures.

Conflicts of Interest

No conflicts of interest were reported.

Acknowledgments

We are very grateful to the children and their parents who were willing to participate in this study. We would like to thank Leo de Raeeve of the Koninklijk Instituut voor Doven en Slechthorenden (Hasselt, Belgium) and Noelle Uilenburg of the Nederlandse Stichting voor het Dove en Slechthorende Kind (Amsterdam, Netherlands) for their continuous support during this study and Dirk-Jan V et for technical support in designing the experiments. We also thank two anonymous reviewers for their helpful comments on earlier versions of this manuscript.

References


Appendix A

Nonword stimuli from Study 1 and Study 2

<table>
<thead>
<tr>
<th>/ɑ/ - /æ/ contrast</th>
<th>/b/-/p/ contrast</th>
</tr>
</thead>
<tbody>
<tr>
<td>/kɑ/ - /kæ/</td>
<td>/byk/ - /pyk/</td>
</tr>
<tr>
<td>/tɑ/ - /tæ/</td>
<td>/bet/ - /pet/</td>
</tr>
<tr>
<td>/tæt/ - /tæt/</td>
<td>/bɛt/ - /pɛt/</td>
</tr>
<tr>
<td>/m/ - /y/ contrast</td>
<td>/f/ - /s/ contrast</td>
</tr>
<tr>
<td>/kɪ/ - /kɨ/</td>
<td>/fɪk/ - /sɪk/</td>
</tr>
<tr>
<td>/ɡɪk/ - /ɡɨk/</td>
<td>/fot/ - /sot/</td>
</tr>
<tr>
<td>/tɪ/ - /tɨ/</td>
<td>/fɛt/ - /sɛt/</td>
</tr>
</tbody>
</table>
Appendix B

/open/-/close/ contrast

/eye/-/chin/ contrast

Non-minimal non-sign pairs in Study 2

Nonsign stimuli from Study 1 and Study 2.