Peer attitudes toward children with cleft (lip and) palate related to speech intelligibility, hypernasality and articulation

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Reference

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Abstract

Background and aims

In addition to anatomical and physiological problems, children with a cleft (lip and) palate (CP±L) often face psychosocial difficulties. A complex interaction between patient and environment may induce these problems. Based on the literature, speech disorders may negatively influence a listener's judgement of a speaker. Therefore, the aim of the present study was to investigate the attitudes of peers toward the speech of children with CP±L.

Method

Sixty-nine typically-developing children (7-12 years, 34 boys, 35 girls) judged audio-recorded speech samples of nine children with CP±L and three control children based on three attitude components, i.e. cognitive, affective and behavioral. A speech intelligibility percentage was determined for each speaker based on transcriptions by 23 naïve adult listeners. Furthermore, two speech-language pathologists perceptually rated the degrees of hypernasality, nasal airflow and articulation errors. A correlation was calculated between the attitude components and the speech intelligibility percentage, and the attitude components and perceptual judgements. Additionally, the possible influence of age and gender of the listeners on their attitudes was explored.

Results

A significantly positive correlation was found between the speech intelligibility percentage and the attitude components: when a child was understood better, more positive attitudes were measured. A significantly negative correlation was found between perceptual judgements and all attitudes components: presence of more hypernasality, nasal airflow or articulation errors resulted in more negative attitudes. Furthermore, boys and younger children seem to have more negative attitudes compared to girls and older children.

Conclusion

This study provides additional evidence that peers show more negative attitudes toward children with more speech disorders due to CP±L. Further research may explore the possible impact of age and gender on attitudes of peers. Intervention should focus on, changing the cognitive, affective and behavioral attitudes of peers in a more positive direction and encouraging the psychosocial development of children with CP±L.

Introduction

Children with a cleft (lip and) palate (CP±L) often face different problems during their development (Kummer, 2008; Peterson-Falzone, Hardin-Jones, & Karnell, 2010). These problems can be directly or indirectly related to their cleft. On the one hand, structural defects can cause feeding, hearing and speech disorders. On the other hand, they may suffer from psychosocial problems, such as symptoms of depression, behavioral problems or teasing by peers (Hunt, Burden, Hepper, Stevenson, & Johnston, 2006; Murray et al., 2010). Based on the literature, 21 to 47 percent of the children with CP±L reported that they have been a victim of teasing or bullying due to speech problems at least once in their life (Havstam, Laakso, Lohmander, & Ringsberg, 2011; Hunt et al., 2006; Noor & Musa, 2007; Semb et al., 2005; Turner, Thomas, Dowell, Rumsey, & Sandy, 1997). This teasing may have a negative impact on the child's self-confidence (Noor & Musa, 2007). Furthermore, Murray et al. (2010) reported that school-aged children with CP±L show more social difficulties based on observations of peer interactions at school. More specifically, they spend more time alone, are less involved in group play and experience more negative interactions with peers than children without cleft. Moreover, internalizing problems, social problems and anxious and withdrawn-depressed behavior were more commonly reported in these children based on teacher reports. Child communication problems largely accounted for these difficulties, especially in children with CP±L (Murray et al., 2010).

The extent to which children with CP±L experience psychosocial difficulties depends on a complex interaction of different influencing factors (Feragen, Særvold, Aukner, & Stock, 2017). The International Classification of Functioning, Disability and Health (ICF) presented by the World Health Organization (WHO) emphasizes the complex interaction between a patient's health condition on the one hand and contextual factors on the other hand (World Health Organization, 2001). In their holistic framework, organic impairment of body structures and functions are connected to implications on the patient's activities and participation as well as to the influence of the person itself and his/her environment (World Health Organization, 2001). One of the most important aspects within these environmental factors are the attitudes of the patient's surrounding people. Positive societal attitudes play an important role in the inclusion of people with disabilities and in limiting the transformation of a functional disability into a more generalized personal, family, social or vocational handicap (Findler, Vilchinsky, & Werner, 2007). These attitudes may be related to a speaker's speech characteristics, such as voice, resonance and articulation, as these often influence a listener's perception and judgement regarding age, health, educational and social status of that speaker (Allard & Williams, 2008). In this light, several studies reported negative stereotyping or misclassification of people with speech disorders, such as voice disorders (Lass, Ruscello, Bradshaw, & Blankenship, 1991; Ma & Yu, 2013) and speech sound disorders (Crowe Hall, 1991; Overby, Carrell, & Bernthal,

2007). Most of these studies investigated the perceptions of adult listeners, such as naïve listeners (Allard & Williams, 2008), teachers (Ma & Yu, 2013; Overby, et al., 2007) and speech-language pathology students (Lass, Ruscello, Pannbacker, Schmitt, & Everly-Myers, 1989; Ma & Yu, 2013). However, the ICF model of the WHO highlights the inclusion of peers' attitudes as an important additional environmental factor in children.

To the best of our knowledge, three studies have focused on the relationship between perceptual ratings of speech in children with CP±L based on audio samples and social acceptance of these children by peers. Different aspects of speech (i.e. nasality, articulation and overall speech intelligibility) were analyzed. The first study focused on the relationship between perceptual ratings of nasality and social acceptance of children with CP±L by peers (Watterson, Mancini, Brancamp, & Lewis, 2013). Seven children with CP±L and hypernasal speech and three children without CP±L or speech problems who served as controls were included. Speech samples based on the reading of an oronasal text passage (i.e. a text passage including oral and nasal phonemes with a distribution comparable to the distribution in spontaneous English speech (Fairbanks, 1960)) were collected. Forty-four normally developing children between 8 and 11 years old rated the degree of hypernasality of each sample. Additionally they completed five social acceptance statements related to the patient on a three-point Likert scale. The authors concluded that increased nasality was associated with decreased social acceptance by peers, and that even mild hypernasality may result in negative consequences for the child with CP±L.

In the second study, Nyberg and Havstam (2016) analyzed how peers describe the speech and communicative participation of children with CP±L. Speech samples based on sentence repetition of ten children with speech disorders due to CP±L were included. Nineteen normally developing 10-year old children described the speech and communicative participation of children with CP±L during focus group sessions. A quantitative content analysis resulted in three main categories of their utterances: (1) description of the speech, (2) thinking about causes and consequences, and (3) emotional reactions and associations. Based on this analysis, the authors concluded that even minor articulation errors were noticed by peers, but that nasality needed to be remarkably present before it was mentioned. This last conclusion is in contrast with the results reported by Watterson et al. (2013). Methodological differences, i.e. qualitative versus quantitative analyses, in which more specific attention was paid to nasality in the study by Watterson et al. (2013) may explain this difference.

The third study explored the relation between speech intelligibility in children with CP±L and social and personal attribute judgements made by peers (Lee, Gibbon, & Spivey, 2017). Speech samples based on sentence repetition of eight children with reduced speech intelligibility due to CP±L and two children without CP±L or speech disorders who served as controls were included.

Ninety normally developing children between 7 and 12 years old judged eight non-speech, social and personal traits in the form of bipolar adjective pairs completed with a neutral statement (e.g. kind – I'm not sure – mean). Additionally, 20 non-expert adult listeners transcribed each speech sample to define a speech intelligibility percentage for each speaker. The authors concluded that a reduced speech intelligibility was related to perceptions by peers of "sickness", "having no friends" and "looking ugly".

Overall, these studies concluded that children with CP±L are less socially accepted by peers if they show more speech problems. However, no study yet has included the most important speech characteristics that can influence speech intelligibility in children with CP±L, i.e. resonance disorders, audible nasal airflow and articulation errors. Therefore, we cannot conclude which speech characteristic has the most impact on the attitudes of peers. Furthermore, Findler et al. (2007) pointed at the multidimensional character of attitudes in which attitudes consist of three different dimensions: a cognitive, affective and behavioral dimension. More specifically, the cognitive dimension focuses on the attribution of, for example, personal and social traits to a person, the affective dimension includes the feelings of a person toward someone else, and the behavioral dimension relates to the behavior of a person toward someone else, e.g. the willingness to help that person. Consequently, they emphasize the need to include all three dimensions in studies regarding people's attitudes toward persons with disabilities. All abovementioned studies mainly focused on the attribution of personal and social traits to speakers with CP±L, which can be related to the cognitive dimension. However, no information is yet available regarding the affective and behavioral attitudes of peers toward children with CP±L.

Furthermore, the influence of age and gender on the attitudes of peers remains unclear. Based on socio-cognitive development research, younger children are more egocentric, meaning that it is more difficult for them to see situations from another person's perspective which may result in more negative attitudes toward peers with disabilities (Alvarez, Ruble, & Bolger, 2001; Werner, Peretz, & Roth, 2015). However, the only study regarding the attitudes of peers toward children with CP±L that included the influence of age reported no conclusive results (Lee et al., 2017). More specifically, the youngest children (7-8 years) assigned the trait "mean" significantly more often than the trait "kind" compared to the older children (9-12 years). On the contrary, they assigned the trait "out-going" more often than the trait "shy". To the best of our knowledge, no studies have explored the influence of gender on attitudes regarding children with CP±L. Based on socio-cognitive development research, girls are, in general, more empathic than boys (Landazabal, 2009; Litvack-Miller, McDougall, & Romney, 1997; Taylor, Eisenberg, Spinrad, Eggum, & Sulik, 2013). Because several correlational studies found a positive significant association between empathy and prosocial behavior (Eisenberg, Eggum, & Di Giunta, 2010; Taylor et al., 2013), this may hypothesize that girls

show more positive attitudes toward peers with disabilities. However no straightforward results are reported in literature (Werner et al., 2015).

Information regarding the attitudes of peers toward children with CP±L can be of interest to compose appropriate interventions, including educational strategies to enhance children's understanding and tolerance of differences and difficulties in other children. Therefore, the aim of this study was to explore the attitudes of 7- to 12-year old children regarding the speech of children with CP±L, including the cognitive, affective and behavioral attitude dimension, by investigating their relation with the level of speech intelligibility, the degree of hypernasality, audible nasal airflow and articulation errors in children with CP±L. Additionally, a possible difference in peer attitudes related to age and gender was investigated. We hypothesized that more negative peer attitudes would be related to more severe speech problems. Furthermore, younger children were suspected to have more negative attitudes compared to older children, whereas girls were hypothesized to have more positive attitudes compared to boys.

Method

This prospective study was conducted according to the World Medical Association Declaration of Helsinki and approved by the institutional review and ethical board of the Ghent University Hospital (EC/2016/1570).

Speakers

Data were collected retrospectively from a database of 73 children with resonance disorders who attended the Ghent University Hospital Craniofacial Centre between 2013 and 2016. The inclusion criteria to participate in this study were to be between 6 and 12 years old, to be a native speaker of Dutch, to live in Flanders (the northern part of Belgium), and to have provided a speech sample consisting of spontaneous speech and sentence repetition. Children suffering from a cold or congestion at the moment of testing or presenting with a pharyngeal flap, learning disabilities greater than mild, dysarthria or dyspraxia were excluded from the study. In total, 35 children fulfilled these criteria. Nine children (5 girls, 4 boys, mean age 7.9 years, SD 0.85) consented to participate. Additionally, three children without speech disorders and without CP±L were included as controls. Based on the composition of the patient group, two girls and one boy, aged between 6 and 12 years (mean age 9.0 years, SD 2.16) old, were invited to participate. They were selected by convenience sampling based on the following in- and exclusion criteria: to be a native speaker of Dutch, to live in Flanders, not to have a cold or congestion at the moment of testing, not to have any speech problem, hearing problem, learning disabilities greater than mild, dysarthria or dyspraxia. This information was collected via a questionnaire completed by their parents. Only three control

children were approached and tested because they all met the in- and exclusion criteria. More information regarding participants' demographics can be found in Table 1.

[Please, insert Table 1 here]

Speech protocol

A speech sample based on spontaneous speech and the repetition of 12 oral and 3 nasal Dutch sentences was available for each child. To be able to define the correct content of the spontaneous speech, the investigator verified if she understood everything correctly by asking the child or by asking the parent to translate samples of unintelligible speech. To create the test samples for the listening protocol, the first 65 syllables of spontaneous speech (i.e. the length of the smallest available sample) were selected after deletion of the interjections of the investigator, resulting in speech samples with a similar length in terms of number of syllables. The selection of only conversational speech for the listening protocol was chosen to provide representative information about the articulation and resonance (Kuehn & Moller, 2000). The available sentences were used to perceptually judge articulation (see perceptual analysis). All samples were video-recorded using a Sony HDR-CX280 camera in a quiet room at the clinical department of Ghent University Hospital. To limit listener bias related to the child's appearance, all samples were converted to audio samples using video converter software (Freemake Video Converter, version 1.1.0.66) at a sampling frequency of 48kHz.

Speech intelligibility

For each sample, a speech intelligibility percentage was determined based on the spontaneous speech sample. Therefore, all samples were presented to 23 non-expert adult listeners (10 men, 13 women, mean age 31.6 years, SD 13.45) in free field by using an amplifier (Dell A525 Multimedia Speaker System). They all were native speakers of Dutch, had no subjective hearing or cognitive problems and had no experience with speech disorders related to CP±L. Moreover, they had no experience in transcribing speech samples. This information was collected by questionnaires. They all gave written informed consent. Listening sessions were organized in small groups of 2 to 12 listeners. Each sample was played in free field through speakers (Dell A525 Multimedia Speaker System), once uninterrupted after which each phrase was played twice with a short pause of five seconds between each repetition. All listeners were asked to write down exactly what they heard. A mean speech intelligibility percentage was determined for each sample based on the ratio of the number of correctly identified words to the total number of produced words.

Perceptual analysis

To verify which specific component of speech attributed to the attitudes of the peers, the degrees of hypernasality, audible nasal airflow and articulation errors were judged by two speech-language pathologists with respectively 6 and 2 years of experience in rating resonance and articulation errors related to CP±L. Additionally, the degree of speech intelligibility was also rated by the two SLPs to compare their results with those of the non-expert adults and peers. Blinded audio samples were provided in a randomized sequence using a standard pair of over-ear headphones (Sennheiser EH150). To verify intra-rater reliability, 33% (4/12) of the samples were repeated. Speech intelligibility, resonance and audible nasal airflow were rated based on spontaneous speech. Speech intelligibility was rated based on four categories (0 = severely distorted, 1 = moderately distorted, 2 = mildly distorted, 3 = normal); hypernasality was rated based on five categories (0 = absent, 1 = borderline, 2 = mild, 3 = moderate, 4 = severe), and the frequency of occurrence of audible nasal airflow was judged on a three-point scale (0 = absent, 1 = occasionally heard, 2 = frequently heard). These categories were all based on the definitions and rating system of the Dutch outcome tool for the perceptual evaluation of speech in patients with CP±L (Bruneel et al., 2019). For the evaluation of articulation, phonetic transcription of the 15 sentences was used. Based on this transcription, cleft-related consonant production errors were identified.

To group these cleft speech characteristics (CSCs), the Dutch version of the CAPS-A framework was used (Bruneel et al., 2019; John, Sell, Sweeney, Harding-Bell, & Williams, 2006; Sell, et al., 2009), resulting in four CSCs categories (anterior oral CSCs, e.g. interdentalisation; posterior oral CSCs, e.g. backing to velar/uvular; non-oral CSCs, e.g. glottal articulation; and passive CSCs, e.g. weak/nasalized consonants). This detailed analysis of CSCs was transformed into a four-point scale to indicate the severity of the articulation errors (John et al., 2006). More specifically, the presence of anterior CSCs in 1 or 2 consonants resulted in a mild distortion, whereas the presence of anterior CSCs in 3 or more consonants resulted in a moderate distortion. For all other CSC categories, 1 or 2 affected consonants resulted in a moderate distortion, whereas 3 or more affected consonants resulted in a severe distortion of the articulation. If no consonants were affected, articulation was considered to be normal.

Listeners

A total of 256 children, aged between 7 and 12 years old, were invited to serve as listeners for this study. They were recruited by convenience sampling in three elementary schools in Flanders. All children's parents were asked to complete a questionnaire to verify in- and exclusion criteria and to give written informed consent. One hundred and seven completed questionnaires and consent

forms were returned. Children were excluded based on the following criteria: native language other than Dutch (*n*=1), hearing problems (*n*=0), neurological or velopharyngeal problems (*n*=0), learning disabilities (*n*=24) and being familiar with someone who was born with a CP±L (*n*=4). Additionally, nine children were not attending school at the moment of testing. As a result, 69 children were included and divided in three age categories: 7-8 years, 9-10 years and 11-12 years. Table 2 provides demographic information about the listeners.

[Please, insert Table 2 here]

Attitudes

To determine the attitudes of typically developing peers toward the speech of patients with CP±L, a questionnaire was developed based on the studies by Nowicki (2006); Werner et al. (2015) and Lee et al. (2017). The questionnaire included the three attitude dimensions, i.e. a cognitive, affective and behavioral component. The cognitive dimension included seven positive (happy, clever, out-going, good looking, healthy, kind, having friends) and seven negative (sad, not clever, shy, ugly, sick, mean, no friends) personality and social traits following Lee et al. (2017). Each child was asked to indicate which traits he/she related to the child of the speech sample. A score was calculated by summing the number of positive (+1) and negative (-1) traits following Werner et al. (2015). The affective attitude dimension was investigated by the following three questions: "How do you feel about… listening to this child?", "…this child asking you to play with him/her?", and "…this child asking you to help him/her?". Following Werner et al. (2015), a three-point Likert scale was used with a visualization of each point by an emoticon and additional description (1 = I do not like it, 2 = I do not mind, 3 = I like it). Finally, the willingness to be in social contact with the child was explored using five questions: "Would you like to… play with the child?", "…help the child?", "…share your toys with the child?", "…invite the child to your birthday party?", and "…share a secret with the child?" The questions were based on the Behavioral Intent Scale, describing increasingly more intimate aspects of childhood friendships (Roberts & Lindsell, 1997). Following Nowicki (2006), a four-point Likert scale was used with the visualization of each point by an emoticon and additional description (1 = no, 2 = maybe no, 3 = maybe yes, 4 = yes).

To verify the perception of speech intelligibility, all children were additionally asked to judge the speech intelligibility of each speaker based on a five-point Likert scale with the visualization of each point by an emoticon and additional description (1 = not understood at all, 2 = very few words understood, 3 = some words understood, 4 = most words understood, 5 = all words clearly understood). Based on this information, a correlation could be determined with the more detailed

speech intelligibility score based on the transcriptions by the adult listeners and the speech intelligibility degree as judged by the experienced SLPs.

All children listened to the spontaneous speech samples in groups of 10 to 20 children in a quiet classroom at the children's school. At the start, the investigator explained each question of the above-described questionnaire after which the children could ask for more clarification. Samples were provided twice in free field using an amplifier (Dell A525 Multimedia Speaker System) in a randomized order across the groups of listeners. After each sample, the children got time to complete the questionnaire individually.

Statistical analysis

IBM SPSS Statistics software version 25.0 (IBM Corp., Armonk, NY) was used for the statistical analysis of the data. Intraclass correlation coefficients (ICCs) were calculated to determine the inter-rater reliability of the speech intelligibility percentages using a two-way mixed model (ICC (3,k)) following the classification of Shrout and Fleiss (1979). To determine the inter- and intra-rater reliability of the perceptual analysis of speech intelligibility judged by the SLPs, hypernasality, audible nasal airflow and articulation, quadratic weighted kappas (Fleiss & Cohen, 1973) were used due to the ordinal character of the variables. Further analyses were based on the perceptual analysis of the SLP with the highest overall intra-rater reliability. Descriptive statistics, more specifically means and standard deviations, were determined for each speaker (i.e. speech intelligibility judged by adult listeners and judged by peers) and per group (i.e. patients with CP±L and controls).

Spearman correlation coefficients were used to investigate the correlation between the speech intelligibility percentage based on the transcriptions made by the adult listeners and the speech intelligibility scores given by the peers, and between the speech intelligibility scores given by the peers and the experienced SLPs. Furthermore, Spearman correlation coefficients were applied to verify the correlations between the three attitude dimension scores and the mean speech intelligibility percentages based on adult transcriptions, between the three attitude dimension scores and the speech intelligibility score given by peers, and between the three attitude dimension scores and the degree of speech intelligibility judged by the SLPs. To verify which specific component of speech attributed to the attitudes of the peers, Spearman correlation coefficients were determined between the three attitude dimensions and the degree of perceived hypernasality, audible nasal airflow and articulation errors as judged by the SLPs. Due to multiple comparisons, a Bonferroni correction was applied to the probability level. A probability level of .008 (.05/6) or less was considered to be significant.

To verify gender distribution of the listeners across age groups, a chi-square test was conducted. Additionally, t-tests were applied to compare age by gender within each age group.

Finally, a two-way analysis of variance (ANOVA), with gender and age group as fixed effects, was applied to investigate the effect of age and gender on the three attitude dimensions of the listeners. Separate analyses were performed for each attitude dimension. When an age effect was found without a gender effect, a post-hoc Scheffé test was applied to determine the significance level of the difference between the age groups. Partial omega square (ω^2p) was used to calculate effect sizes. Effect sizes were interpreted following Field (2012), more specifically ω^2p of .01, .06, .14 to indicate small, medium and large effects respectively. A probability level of .05 or less was considered to be significant for these analyses.

Results

Speech intelligibility

Reliability. Based on the guidelines by Cicchetti (1994), excellent agreement was found for the speech intelligibility percentages rated by the non-expert adult listeners. A single measures ICC of .863 (95%CI .751-.949) and an average measures ICC of .993 (95% CI .986-.998) were found. Regarding the ratings by the SLPs, good inter-rater reliability was found (κ = .67), based on the guidelines by Altman (1991). Furthermore, very good intra-rater reliability was found (rater 1: κ = .92; rater 2: κ = 1.00). Further analyses were based on the perceptual analysis of the SLP with the highest overall intra-rater reliability which was the SLP with six years of experience (see also reliability perceptual analysis).

Descriptive statistics. Supplementary Table 1 provides the means and standard deviations of the speech intelligibility percentages judged by the non-expert adult listeners and the speech intelligibility scores judged by the typically developing peers per speaker. Additionally, the degree of speech intelligibility as judged by the first SLP is provided per speaker. Regarding the speech intelligibility percentages based on adult transcriptions, an overall mean of 52.0% (SD 19.79, range 20-78%) was found for the speakers with CP±L and an overall mean of 94.7% (SD 3.22, range 91-97%) for the control speakers without speech disorders or CP±L. For the speech intelligibility scores given by peers, an overall mean of 2.2 (SD .59, range 1.4-3.2) was found for the speakers with CP±L and an overall mean of 4.1 (SD .44, range 3.6-4.4) for the control speakers.

Correlations. A strong and statistically significant positive correlation was found between the speech intelligibility percentages based on the transcriptions by the adult listeners and the speech intelligibility score given by the typically developing peers (r = .91, $p < .001$), and between the degree of speech intelligibility judged by the first SLP and the scores given by the typically developing peers (r = .87, $p < .001$). These results indicate a similar perception of severity of the speech disorder despite the different methodological procedures and the different amount of experience in rating speech intelligibility.

Perceptual analysis

Reliability. Based on the guidelines by Altman (1991), moderate to very good intra-rater reliability was found in both raters for the perceptual judgements of hypernasality (rater 1: κ = .95; rater 2: κ = .97), audible nasal airflow (rater 1: κ = .86; rater 2: κ = .50) and articulation (rater 1: κ = 1.00; rater 2: κ = 1.00). Furthermore, moderate to very good inter-rater reliability was found for the judgements of hypernasality (κ = .73), audible nasal airflow (κ = .58) and the degree of articulation errors (κ = .81). Further analyses were based on the perceptual analysis of the SLP with the highest intra-rater reliability which was the SLP with 6 years of experience.

Descriptive statistics. Supplementary Table 1 provides the results per speaker of the perceptual analysis by the first SLP with 6 years of experience. A range of hypernasality, audible nasal airflow and articulation distortion was represented by the samples of the 12 speakers.

Attitudes

Descriptive statistics. Supplementary Table 1 provides the mean results and standard deviations per speaker for the three attitude dimensions as judged by the normally developing peers.

Correlations. Except for the behavioral attitude dimension, strong and statistically significant positive correlations were found between the attitude dimensions and the mean speech intelligibility percentages based on adult transcriptions, between the attitude dimensions and the mean speech intelligibility scores given by peers, and between the attitude dimensions and the degree of speech intelligibility as judged by the first SLP (Table 3 and supplementary Figures 1, 2 and 3). The better the speech intelligibility of a speaker, the more positive the attitudes of the peers were toward the speaker. Furthermore, strong and statistically significant negative correlations were found between the three attitude dimensions and the perceptual judgements regarding hypernasality, audible nasal airflow and articulation errors (Table 3). The more severe the hypernasality, audible nasal airflow or articulation errors of a speaker, the more negative the attitudes of the peers were toward the speaker. Although all correlations were significant, slightly stronger correlations were found for the variable 'hypernasality' compared to the other perceptual variables.

[Please, insert Table 3 here]

Influence by age and gender of the listeners.

A Chi-square test showed no statistically significant difference for gender distribution across age groups (X^2=2.317, p=0.311). Additionally, no statistically significant differences were found for age between boys and girls in each age group ($p > .05$, Table 2).

Tables 4, 5 and 6 present the results of the two-way ANOVA of the cognitive, affective and behavioral dimension respectively. Based on a two-way ANOVA, no interaction effects between age and gender were found for the cognitive (F (2,63) = .270, p = .764) and affective dimension (F (2,56) = 2.108, p= .131). Nevertheless, significant main effects for age and gender were found for both dimensions. Main effects of age with large effect sizes were found for both the cognitive (F (2,65) = 6.034, p = .004, ω^2p = .129) and affective dimension (F (2,58) = 5.646, p = .006, ω^2p = .132). Post-hoc Scheffé tests showed that the youngest children (7-8 years) attributed significantly lower scores to the speakers compared to the oldest children (11-12 years) in both the cognitive (p = .012) and affective dimension (p = .025). Additionally, girls attributed significantly higher scores with medium to high effect sizes to the speakers in both the cognitive (F (1,65) = 6.375, p = .014, ω^2p = .074) and affective dimension (F (1,58) = 9.650, p = .003, ω^2p = .126), representing more positive attitudes. Regarding the behavioral dimension, a significant interaction effect between age and gender with large effect size was found based on a two-way ANOVA (F (2,57) = 6.666, p = .003, ω^2p = .159). Post-hoc Scheffé tests showed a significant difference between the scores given by the youngest boys (7-8 years) compared to all other groups (boys 9-10 years, p = .025; boys 11-12 years, p = .044; girls 7-8 years, p = .003; girls 9-10 years, p = .045; girls 11-12 years, p = .042), in which the youngest boys attributed significantly lower scores to the speakers, representing more negative attitudes.

[Please, insert Tables 4, 5 and 6 here]

Discussion

This study explored the attitudes of 7-to-12 years old typically developing peers toward children with speech disorders related to CP±L. More specifically, the relationships were investigated between the cognitive, affective and behavioral attitude dimensions of peers and the speech intelligibility, the degree of hypernasality, audible nasal airflow and articulation errors in children with CP±L. Additionally, possible differences in attitudes related to age and gender of the peers were explored.

High and statistically significant correlations were found between the cognitive and affective attitude dimensions and speech intelligibility, whether the intelligibility was determined by a transcription of a continuous speech sample by naïve adult listeners, on a five-point Likert scale by the peers themselves or by an experienced SLP. The more intelligible a speaker was judged, the more positive attitudes the peers had toward the speaker. This finding is comparable with the results reported by Lee et al. (2017), who found a significant positive association between speech intelligibility determined by a similar transcription procedure by naïve adult listeners and several

personal and social traits that peers attributed to speakers with CP±L. The same traits were included in the cognitive dimension in the present study, although the method of presentation differed (i.e. bipolar adjective pairs (Lee et al., 2017) vs. selecting traits related to the speaker in the current study). Despite this methodological difference, comparable results were found. For the behavioral attitude dimension, however, a significant correlation was found only with the speech intelligibility judged by peers.

For all explored variables that often determine speech intelligibility in children with CP±L, more specifically hypernasality, audible nasal airflow and articulation errors, higher degrees of disturbance were significantly related to more negative attitudes in all attitude dimensions. However, correlation coefficients for hypernasality were somewhat higher compared to those of audible nasal airflow and articulation errors. This may indicate that the degree of perceived hypernasality contributes the most to the peers' attitudes toward children with CP±L, although the influence of audible nasal airflow and articulation errors cannot be neglected. To confirm this hypothesis, further research needs to include speech samples of separate groups of children with only hypernasality, articulation errors and, if possible, audible nasal airflow.

Despite the limited literature regarding peers' attitudes toward children with CP±L, some of the present results can be compared with earlier studies. Specifically for hypernasality, the present results are comparable with those of Watterson et al. (2013) who found a decreased social acceptance based on attributed personal and social traits by peers when the speaker presented with more hypernasality. A qualitative study based on focus groups with 10-year-old peers discussing audio samples of children with articulation and resonance disorders related to CP±L by Nyberg and Havstam (2016) also reported the attribution of negative personal and social traits and decreased social acceptance to children with CP±L. However, they indicated that mild hypernasality was often not noticed by peers. The high correlations on the cognitive dimension in the present study, however, are more similar to the results of Watterson et al. (2013) in which peers show more negative attitudes toward children with mild to moderate hypernasality compared to toward children without hypernasality. On the other hand, Nyberg and Havstam (2016) reported that even minor articulation errors were noted by peers. This is in line with the results of the present study, in which a significant correlation was found between the cognitive attitude dimension and the degree of articulation errors. No study yet reported on the possible influence of audible nasal airflow on peers' attitudes toward children with CP±L. Because none of the reported studies regarding attitudes of peers toward children with CP±L included an affective and behavioral attitude dimension, no comparison of the current results is possible. However, observations of peer interactions at school by Murray et al. (2010) found that school-aged children with CP±L spend more time alone, are less involved in group play and experience more negative interactions with peers than children without

CP±L. This is in line with the more negative behavioral attitudes of peers toward children with CP±L in the present study.

The present results may be indicative for the development of prejudices and stereotypes in peers toward children with CP±L. The negative attitudes may result in a lower estimate of the capacities of these children (i.e. cognitive dimension), showing less empathy or affection to these children (i.e. affective dimension) and having fewer social contacts and interaction with these children or even teasing and bullying (i.e. behavioral dimension). This may result in fewer opportunities to build strong relationships or friendships with peers (Lee et al., 2017; Murray et al., 2010), one of the important factors in the development of self-confidence, socio-emotional and socio-cognitive skills in childhood (Gallagher, 1993; Langevin, 2009), which may in turn have a negative impact on psychosocial functioning, education and employment (Havstam et al., 2011; Hunt et al., 2006; Lee et al., 2017). However, disordered speech and resonance are not always associated with self-reported negative social experiences or teasing (Feragen et al., 2017). As stressed by the ICF model of the WHO, a complex interaction between personal and environmental factors influences a child's functioning, activities, participation and perception in addition to the existing disability (e.g. disordered speech and resonance due to CP±L) (Neumann & Romonath, 2012; World Health Organization, 2001). Although peers' attitudes are an important environmental factor, Murray et al. (2010) found that effective linguistic and pragmatic communication skills in children with CP±L may have a positive effect on the children's social interactions and socio-emotional development. Another important personal factor in social interactions is the perception of teasing. The perception of teasing is related to greater psychosocial problems (Hunt et al., 2006). However, how teasing is perceived depends on the psychosocial vulnerability or strength of the child with CP±L (Feragen et al., 2017). Furthermore, appearance, behavior and social skills are mentioned as possible personal factors that may influence relationships with peers (Hunt et al., 2006; Nyberg & Havstam, 2016). Nevertheless, it remains a complex interaction in which cause and effect are difficult to define, resulting in the need for intervention in both parties.

A significant influence of age and gender with medium to large effect sizes was currently found on the attitudes of peers toward children with CP±L. Overall, the youngest children showed more negative attitudes compared to the oldest in all three attitude dimensions. This confirms our hypothesis based on the socio-cognitive development theory. This theory presumes that young children reason according to their personal experiences and concrete actions with objects, persons and events (Littlefield-Cook, Cook, Berk, & Bee, 2005). As a result, these children may prefer peers with similar traits, e.g. normal intelligibility, and attribute more negative attitudes to children who are different. Moreover, children under the age of 8 years old tend to attribute a more global categorization of peers based on evaluative judgements, such as good versus bad, of one outstanding

feature (Alvarez et al., 2001; Nowicki, 2006). In the present study, speakers with poor intelligibility may have been overall categorized as bad by the younger children, which may explain the significant differences in attitudes between the youngest and oldest age groups. In the literature, studies that include a wider age range with younger children (4-6 years) often report similar results in which younger children show more negative attitudes toward children with disabilities (Nowicki, 2006; Werner et al., 2015). However, a meta-analysis by Nowicki and Sandieson (2002) showed no conclusive evidence for an age trend in attitudes toward persons with physical or intellectual disabilities. In children with CP±L, Lee et al. (2017) also did not find straightforward results regarding the influence of age on attitudes of peers toward children with CP±L, in which some negative personal and social traits were more assigned by the youngest children whereas other negative traits were more assigned by the oldest. Consequently, further research is necessary to confirm the present findings. Regarding the influence of gender, boys had significantly more negative attitudes compared to girls in all three attitude dimensions. This is in line with our hypothesis based on socio-cognitive development research. However, the current results may have been influenced by the unequal distribution of gender in the speakers (58% girls, 42% boys), which is a limitation of the present study. Because children prefer to associate with their own gender (Nowicki, 2006; Sippola, Bukowski, & Noll, 1997), this may explain the more positive attitudes in girls. Furthermore, Nowicki (2006) found that girls overall attributed more positive traits to children whether they present with a physical or intellectual disability or not. Further research is necessary to confirm these hypotheses, including children with CP±L and taking gender matching into account. On the other hand, the inconclusive results regarding age and gender in the literature may also indicate that they are not directly related to attitudes. Therefore, it could be interesting to investigate the correlation between the socio-cognitive or psychosocial development level of peers and their attitudes toward children with disabilities, which may be influenced by age and gender of the listeners but also by other characteristics such as their maturity and education.

As supposed by other authors, the applied methods may have had an influence on the results. First, the speech samples were based on spontaneous speech of children with CP±L without content control. Children talked about school, leisure and sport activities. The content of the speech sample may have influenced the attitudes of the listeners. In further research, controlled content of the speech sample, e.g. retelling a standardized story based on pictures, may exclude this influencing factor. Second, only a limited number of answer possibilities per rating scale were included due to the age of the listeners. Although this choice was based on previous attitude studies in children (Nowicki, 2006; Werner et al., 2015), this may have prevented the children from responding more nuancedly. Third, only audio samples were provided to the listeners. In further research, it would be interesting to include additional video samples to compare the possible influence of appearance on

the attitudes of peers, especially toward children who were born with a cleft lip in addition to a cleft palate. Fourth, none of the included peers was ever exposed to an individual with CP±L. Several studies demonstrated that having contact with an individual with a disability may result in having more positive attitudes toward those persons (see Blancher and Goodwyn (2016) for an overview). Whether exposure to children with CP±L would result in more positive attitudes in peers, e.g. when a child with CP±L is attending the same class, is subject for further research. Finally, the inclusion of only nine speech samples of children with CP±L can be considered as a limitation of this study. Although this sample size echoed the previous studies regarding peers' attitudes and limited the task load of the young listeners during the listening experiment, the full spectrum of speech intelligibility in children with CP±L may not have been covered by the included speech samples. As a result, conclusions should be interpreted with care.

Nevertheless, the present results are in line with the literature and therefore emphasize the need for intervention in order to change negative attitudes toward children with CP±L. As mentioned by Murray et al. (2010), especially the school context is often challenging for these children, making it the ideal environment to implement intervention programs related to attitude change in peers. As mentioned above, information on attitudes and exposure to an individual with a disability may result in more positive attitudes. Specifically related to children with CP±L, Blancher and Goodwyn (2016) found that providing information in combination with having contact with an adult with CP±L increased the cognitive attitudes in children between 9 and 11 years old. However, no effect was found for the affective and behavioral attitude dimension. This result was explained by Edwards' match hypothesis (Edwards, 1990), saying that only those attitudinal components will be influenced that are included in the intervention. As more negative attitudes related to less intelligible and more hypernasal speech were found for all three attitude dimensions in the present study, it will be necessary to address those three dimensions in the development of a specific intervention program. In addition to information and exposure, this may include, for example, exercises to improve empathy among peers that differ (Batson & Ahmad, 2009) and role-play and simulated learning to discuss interactions and behavior as suggested by Blancher and Goodwyn (2016). Additionally, it appears to be important to address attitudes over an extended period to effect real change (Kathard et al., 2014; Langevin & Prasad, 2012). To the best of our knowledge, no literature about the effectiveness of a specific intervention program to influence attitudes of peers toward children with CP±L is yet available. Whether these intervention programs should be age- or gender-related given the current results, is subject for further research.

In conclusion, this study provided additional evidence that peers show more negative attitudes toward children with less intelligible and more hypernasal speech due to cleft lip and palate. This study was the first to include all three attitude dimensions, i.e. the cognitive, affective

and behavioral dimension. Highly significant correlations were found for all three dimensions between the attitudes of peers and the speech intelligibility, the degree of hypernasality, audible nasal airflow and articulation errors in children with CP±L. Further research is necessary to explore the possible impact of age and gender on attitudes of peers and whether those influencing factors should be included when developing and evaluating specific intervention programs. Due to the complex interaction of environmental and internal factors in building friendships with peers, intervention should focus on both, changing the cognitive, affective and behavioral attitudes of peers in a more positive direction and encouraging the psychosocial development of children with CP±L.

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Tables

Table 1. Speaker's demographic information and medical history.

Speaker	Gender	Age (year; months)	Diagnosis
1	F	10;10	control
2	M	9;6	control
3	F	8;11	control
4	F	8;1	BCLP
5	M	10;6	BCLP
6	M	6;3	UCLP
7	F	7;3	CPO
8	F	8;5	CPO
9	M	7;8	CPO
10	F	10;2	CPO
11	F	10;1	CPO
12	M	6;7	CPO

BCLP: bilateral cleft lip and palate; UCLP: unilateral cleft lip and palate; CPO: cleft palate only

Table 2. Listener's demographic information: age and gender.

	Total			***Boys***			***Girls***			***Comparison of age by gender; Independent student's t-test***	
Age group	**N**	**Mean age (in years)**	**SD**	**N**	**Mean age (in years)**	**SD**	**N**	**Mean age (in years)**	**SD**	***t***	***p***
7-8 years	30	7.2	.55	14	8.0	.63	16	7.8	.48	1.001	.326
9-10 years	22	10.0	.56	9	9.9	.48	13	10.1	.61	-.892	.383
11-12 years	17	11.6	.35	11	11.7	.31	6	11.6	.47	.302	.767
Total	69	9.4	1.63	34	9.7	1.68	35	9.2	1.57	1.130	.263

*Independent student's t-test, $p < .05$

Table 3. Spearman correlation coefficients and significance values representing the correlations between the three attitude dimensions and the speech intelligibility percentage based on transcriptions made by adults, the speech intelligibility score judged by peers, the speech intelligibility degree judged by a speech-language pathologist, and perceived hypernasality, audible nasal airflow and articulation judged by a speech-language pathologist.

	Cognitive dimension	**Affective dimension**	**Behavioral dimension**
Speech intelligibility percentage - adults	r = .80* *p* = .002	r = .77* *p* = .003	r = .70 *p* = .012
Speech intelligibility score - peers	r = .90* *p* < .001	r = .92* *p* < .001	r = .82* *p* = .001
Speech intelligibility degree – SLP	r = .75* *p* = .005	r = .73* *p* = .007	r = .68 *p* = .016
Hypernasality	r = -.74* *p* < .001	r = -.66* *p* < .001	r = -.64* *p* < .001
Audible nasal airflow	r = -.57* *p* < .001	r = -.48* *p* = .001	r = -.49* *p* < .001
Articulation	r = -.65* *p* < .001	r = -.51* *p* < .001	r = -.48* *p* = .001

*Spearman correlation with Bonferroni correction, $p \leq .008$

Table 4. *Cognitive attitudes.* Descriptive statistics and ANOVA results regarding the influence by gender and age of the listeners on their cognitive attitudes toward children with speech disorders related to cleft palate.

Descriptive statistics				
	Total *Mean ratings (SD), range -7 to +7*	*7-8 years* *Mean ratings (SD), range -7 to +7*	*9-10 years* *Mean ratings (SD), range -7 to +7*	*11-12 years* *Mean ratings (SD), range -7 to +7*
Total	.33 (1.89)	-.25 (1.99)[a]	.28 (1.81)	1.40 (1.41)[a]
Boys	-.10 (2.14)	-1.00 (2.18)	-.19 (2.44)	1.13 (1.14)
Girls	.74 (1.54)	.42 (1.58)	.62 (1.20)	1.89 (1.82)
ANOVA results				
Predictor		***F***	***p***	ω^2_p
GENDER		6.375	.014*	.074
AGE		6.034	.004*	.129
GENDER x AGE		.270	.764	N.A.

*****Two-way analysis of variance, $p \leq .05$

[a]Post-hoc Scheffé test, $p \leq .05$; significant difference between age groups 7-8y and 11-12y, $p = .012$.

Table 5. *Affective attitudes.* Descriptive statistics and ANOVA results regarding the influence by gender and age of the listeners on their affective attitudes toward children with speech disorders related to cleft palate.

Descriptive statistics				
	Total *Mean ratings (SD), range 1-3*	*7-8 years* *Mean ratings (SD), range 1-3*	*9-10 years* *Mean ratings (SD), range 1-3*	*11-12 years* *Mean ratings (SD), range 1-3*
Total	2.19 (.44)	2.02 (.50)[a]	2.28 (.35)	2.36 (.36)[a]
Boys	2.05 (.43)	1.72 (.34)	2.18 (.38)	2.33 (.30)
Girls	2.33 (.42)	2.27 (.49)	2.36 (.32)	2.43 (.48)
ANOVA results				
Predictor		***F***	***p***	ω^2_p
GENDER		9.650	.003*	.126
AGE		5.646	.006*	.132
GENDER x AGE		2.108	.131	.036

*****Two-way analysis of variance, $p \leq .05$

[a]Post-hoc Scheffé test, $p \leq .05$; significant difference between age groups 7-8y and 11-12y, $p = .025$.

Table 6. *Behavioral attitudes.* Descriptive statistics and ANOVA results regarding influence by gender and age of the listeners on their behavioral attitudes toward children with speech disorders related to cleft palate.

Descriptive statistics				
	Total *Mean ratings (SD), range 1-4*	*7-8 years* *Mean ratings (SD), range 1-4*	*9-10 years* *Mean ratings (SD), range 1-4*	*11-12 years* *Mean ratings (SD), range 1-4*
Total	2.51 (.65)	2.31 (.83)	2.62 (.49)	2.68 (.45)
Boys	2.32 (.63)	1.78 (.48)[a]	2.71 (.56)	2.62 (.35)
Girls	2.70 (.63)	2.80 (.79)	2.57 (.44)	2.77 (.62)
ANOVA results				
Predictor		***F***	***p***	ω^2_p
GENDER		5.430	.023*	.070
AGE		3.271	.045*	.070
GENDER x AGE		6.666	.003*	.159

*****Two-way analysis of variance, $p \leq .05$

[a]Post-hoc Scheffé test, $p \leq .05$; significant difference between youngest boys (7-8y) and all other groups, $p \leq .05$.

Supplementary Table 1. Mean speech intelligibility percentages (M) and standard deviations (SD) per speaker based on judgments of 23 non-expert adult listeners, mean speech intelligibility and attitude dimension scores and standard deviations per speaker based on judgments of 69 normally developing peers, and degree of speech intelligibility, perception of hypernasality, audible nasal airflow and articulation based on judgments of a speech-language pathologist.

	S1*	**S2***	**S3***	**S4**	**S5**	**S6**	**S7**	**S8**	**S9**	**S10**	**S11**	**S12**
	M (SD)	**M (SD)**	**M (SD)**	**M (SD)**	**M (SD)**	**M (SD)**	**M (SD)**	**M (SD)**	**M (SD)**	**M (SD)**	**M (SD)**	**M (SD)**
Speech intelligibility %	96 (3.37)	91 (3.45)	97 (2.84)	55(12.4)	77 (9.22)	20 (11.6)	49 (10.4)	26 (9.26)	47 (14.4)	55 (12.1)	78 (8.41)	61 (15.1)
Speech intelligibility score (1-5)	4.35 (.98)	3.59 (1.41)	4.34 (1.11)	2.30 (1.02)	1.88 (1.09)	1.38 (.62)	2.28 (1.06)	1.58 (.81)	1.68 (1.00)	2.23 (1.06)	3.20 (1.39)	2.83 (1.21)
Degree of speech intelligibility	Normal	Normal	Normal	Severe	Moderate	Severe	Mild	Severe	Severe	Severe	Mild	Mild
Degree of hypernasality	Absent	Absent	Absent	Mild	Moderate	Severe	Mild	Severe	Severe	Mild	Moderate	Borderline
Degree of audible nasal airflow	Absent	Absent	Absent	Occasionally	Frequently	Absent	Occasionally	Absent	Frequently	Absent	Frequently	Absent
Degree of articulation errors	Normal	Normal	Normal	Severe	Mild	Severe	Normal	Severe	Severe	Moderate	Normal	Moderate
Cognitive dimension (-7 to +7)	3.58 (2.70)	2.00 (3.58)	3.26 (3.34)	-0.51 (2.46)	-1.58 (3.17)	-1.55 (2.63)	-.81 (3.31)	-.72 (3.13)	-1.70 (3.07)	-.43 (3.07)	1.55 (3.41)	.84 (3.28)
Affective dimension (1-3)	2.66 (.47)	2.43 (.53)	2.54 (.57)	2.20 (.53)	1.93 (.67)	1.97 (.50)	2.02 (.61)	2.04 (.65)	1.98 (.67)	2.07 (.66)	2.25 (.65)	2.23 (.69)
Behavioral dimension (1-4)	2.98 (.81)	2.77 (.85)	2.85 (.85)	2.56 (.66)	2.15 (.91)	2.36 (.76)	2.24 (.84)	2.28 (.92)	2.14 (.83)	2.31 (.89)	2.53 (.91)	2.58 (.96)

*speaker without speech disorders and without cleft (lip and) palate

Figures

Supplementary Figure 1. Scatter plot representing the correlations between the three attitude dimensions and the speech intelligibility percentage based on transcriptions made by non-expert adult listeners. Filled markers represent the children without speech disorders and without cleft (lip and) palate.

Supplementary Figure 2. Scatter plot representing the correlations between the three attitude dimensions and the speech intelligibility score judged by peers ranging from 1 (not understood at all) to 5 (all words clearly understood). Filled markers represent the children without speech disorders and without cleft (lip and) palate.

Supplementary Figure 3. Scatter plot representing the correlations between the three attitude dimensions and the speech intelligibility score judged by a speech-language pathologist ranging from 0 (severely disturbed) to 3 (normal). Filled markers represent the children without speech disorders and without cleft (lip and) palate.