



Speech intelligibility after repair of cleft lip and palate

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Abstract

Background: Intelligibility refers to understandability of speech; and lack of it can negatively affect children's overall communication effectiveness. Children with repaired cleft lip and/or cleft palate (CL/P) may experience poor speech intelligibility. This study aimed at evaluating speech intelligibility in children with repaired CL/P who had not been referred to speech-language pathology clinics for early intervention.

Methods: Sixty-four monolingual Persian-speaking children, 32 children with repaired CL/P, and 32 controls aged 3 to 5 years participated in this survey. Their speech intelligibility was evaluated through the Persian Speech Intelligibility Test and was normalized on children 3 to 5 years. Each speech sample was heard by a speech and language pathologist (SLP), as well as 2 nonprofessional listeners. Two objective measures of speech intelligibility including Percentage of Consonants Correct (PCC) and Percentage of Intelligible Words (PIW) were used in this research.

Results: Children with CL/P were significantly outperformed by their peers in PCC ($p=0.0001$) and PIW ($p=0.0001$). More than half of the case group had compensatory errors and 40.6% had obligatory errors. The PCC and the PIW were statistically different in children with different rates of hyper nasality ($p=0.001$).

Conclusion: Speech intelligibility of children with CL/P is impaired due to their articulation disorders (obligatory and compensatory errors). This survey documents the necessity for speech therapy for increasing speech intelligibility in this population.

Keywords: Speech Intelligibility, Percentage Intelligible Words, Percentage Consonants Correct, Cleft Lip and/or Palate

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Introduction

The term "speech intelligibility" refers to speech clarity of a speaker's output that a listener can readily understand. Estimating percentages of intelligibility is one of the measurements that can be used by SLPs. The speech materials used for this purpose may consist of single words, sentences, and spontaneous speech (1). Score of speech intelligibility for normal English-speaking children has been reported to be 75% by age 3, 90% by age 4, and 100% by age 5 (2). Similarly, based on the studies conducted on Persian speaking children, the speech intelligibility scores of 3 and 4-year-old children are 83% to 92%, and 92% to 96%, respectively (3).

Cleft lip and/ or cleft palate (CL/P) is one of the most prevalent congenital craniofacial malformations (4), with a prevalence of 1 per 1000 births in America (5). Kianifar et al. explored the incidence and related factors of CL/P among live births in Mashhad, Northeastern Iran. They reported that the overall incidence of CL/P was 1.9 per 1000 live births (6). The most (50%) prevalent type of cleft was cleft lip and palate (CLP), followed by cleft lip (35.2%), and cleft palate (14.8%). The prevalence of clefts was more common in male than in female births (male/female ratio was 3:2) (7).

Cleft lip/palate repair is a surgical procedure requiring

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↑What is "already known" in this topic:

We know that the surgery solely cannot solve the intelligibility problem in children with cleft lip and palate, and a combination of surgery as well as SLP services is recommended.

→What this article adds:

All participants with cleft lip and palate received the surgery but not the SLP services. The relationship between Percentage Intelligible Words and Percentage Consonants Correct showed that those children who could produce more words with correct consonants had higher speech intelligibility. More than 50% of children had compensatory error that could be treated by speech therapy.

a variety of techniques to restore function and to restore a more normal appearance. However, Bicknell et al. suggested that this repair may not always lead to a normal function of lip and palate (8). In other words, these children usually have speech disorders even after the surgery. As a case in point, Sell et al. conducted a comprehensive survey of speech outcomes in children born with unilateral cleft lip and palate and reported speech unintelligibility, hypernasality, and consonants error in these children. In this study, almost two-thirds of children had undergone speech therapy (9).

Speech disorders are strongly associated with reduced intelligibility. As a case in point, McWilliams concluded that there is a direct relationship between speech intelligibility and severity of nasality and articulation errors (10). Similarly, Hun examined the relation between speech intelligibility and percentage of consonants correct (PCC) and concluded that speech intelligibility is influenced by PCC (11).

Lohmender and Christian longitudinally studied 20 children aged 3 to 7 years with CL/P and compared them with typically developing children. They concluded that the PCC and the percentage of correct place of articulation were significantly lower in children with CL/P than in normal children (12).

Speech sound errors in children with cleft palate are classified into 2 types: obligatory and compensatory. Obligatory errors are caused by structural disturbances (hypernasality), but compensatory errors are misarticulations due to the misplacement of articulators which have been learned during speech development (including glottal stops, pharyngeal fricatives, and pharyngeal plosives) (13).

Based on the study conducted by Hardin-Jones and Jones, 37% of preschool children had obligatory errors and 25% had compensatory errors (14). The association between middle ear disease and cleft palate is well documented. Although the tendency to develop middle ear disease will be reduced by early palatal surgery, it is necessary to evaluate and follow up hearing performance of children with cleft palate every 6 months. Ibrahim et al. studied speech and hearing outcomes in children with CL/P. They reported that only 19% of participants had normal speech intelligibility rating and normal hearing bilaterally (14). Similarly, Schönweiler et al. suggested that the hearing status has a profound effect on the speech and language abilities in CLP patients (15).

Considering the fact that children with CL/P have a variety of speech and language disorders, speech-language pathology services are required to improve the functional communication in this population. One of the main goals in speech therapy is to increase speech intelligibility. Lockhart suggested that almost 50% of children with CL/P will need speech and language therapy (16). Unfortunately, there are children who are not referred to SLP clinics after surgery.

The main objective in the current study was to evaluate speech intelligibility in children with repaired CL/P who had not been referred to SLP clinics for early intervention.

Methods

This cross-sectional study was conducted on children aged 3 to 5 years. The speech of 32 children with repaired CL/P was compared with 32 age- and sex-matched control children (22 girls and 42 boys). All participants were monolingual and spoke Persian as their first and main language. Children with CL/P had been referred to hospitals and SLP clinics in Mashhad, Iran, from September to November 2015, and they were recruited from these centers. This study was approved by the ethics committee of Varastegan Institute of Medical Sciences.

All participants were examined in the audiology clinic of Ghaem hospital by an audiologist who conducted pure tone audiometry testing (PTA) using the CA86 clinical audiometer and Telefonix TBH39P headphones. The examinations were done at the frequencies of 250 Hz, 500 Hz, 1000Hz, 2000Hz, 4000Hz, and 8000Hz in both ears. Thresholds higher than 15dB HL were considered as a hearing loss (17). Hearing assessment revealed that children with CL/P had a mild hearing loss between 25 to 40dB, which was mostly seen as a conductive hearing loss. It should be mentioned that none of the children presented mixed hearing loss.

Based on the Age & Stage Questionnaire, the control group's scores on communication, fine motor, gross motor, personal, social, and problem-solving skills were within normal limits. Each domain was scored separately, and the scores were compared with the screening cutoff scores of each domain (18). The children had no history of neurological problems, seizures, brain damage, or any other disorders and they did not have any symptoms of movement delay. All children were evaluated by a speech-language therapist with respect to oral assessment, speech intelligibility assessment, and assessment of articulation and resonance.

Speech intelligibility assessment was performed using the Persian Speech Intelligibility Test (single-word) validated for 3 to 5 years-old children (19). All children were evaluated in an appropriate room with minimum noise and sufficient light.

The test pictures were displayed to the children via a laptop (Dell-INSPIRON- 6400) with a 17" screen. The children were asked to name the test pictures verbally. The speech samples of children were recorded by the Audacity software using the headset microphone so that the microphone-to-mouth distance was 10 centimeters. The native Persian-speaking listeners were trained for writing the orthographical transcription of speech samples. The listeners heard the recorded speech samples of children and transcribed every word they could understand. Each speech sample was heard by 2 listeners. Furthermore, an SLP transcribed speech samples of children were used later for the PCC calculation. Two methods, PIW and PCC, were used to measure speech intelligibility. The PCC was calculated by dividing the total number of correct consonants by the total number of consonant (20).

Likewise, the PIW was obtained through the division of the total number of intelligible words by the total number of word targets (21). The assessment of articula-

tion and resonance was performed using the cleft palate speech assessment test based on the universal parameters system in Persian (22), and oral assessment was performed via the Oral Speech Motor Control Protocol in Farsi Speaking Children (23). To examine the interrater point-to-point agreement reliability (24), the percentage agreement was calculated. The language samples of 20% of children were randomly selected, re-transcribed by another trained listener, and the percent agreement was determined (24). The result indicated 90% interrater agreement on words, demonstrating an acceptable level of agreement (24).

Statistical analysis was conducted using SPSS 21. A Kolmogorov–Smirnov test at $P \geq 0.05$ was conducted, which indicated that the data were not normally distributed, and hence, nonparametric testing was implemented. Spearman's correlation coefficient was used to study the correlation between the 2 intelligibility variables (PCC and PIW). The Mann–Whitney U test was conducted to evaluate the effect of gender on the PCC and PIW variables. Moreover, the Kruskal–Wallis test was performed to compare the speech intelligibility in 4 groups of children with different rates of hypernasality (normal, mild, moderate, and severe hypernasality). Statistical significance level was set at $p < 0.05$.

Results

Table 1 demonstrates the different types of CL/P in children. Almost 90% of the children with CL/P had mild to severe hypernasality (mild= 34.4%, moderate= 34.4, and severe= 21.9). Descriptive analysis of the variables is presented in Table 2. The mean of PCC and PIW in children with CL/P was 61.77 % and 59.68%, respectively. The results of the Mann–Whitney U test revealed a significant difference between children with CL/P and the normal group in the mean scores of PCC ($p = 0.0001$) and PIW ($p = 0.0001$). The results of the Spearman coefficient ($\rho = 0.91$) revealed a significant relationship between PCC and the PIW in children with CL/P ($p = 0.0001$) (Fig. 1).

The results revealed a significant relationship between the severity of hypernasality and speech intelligibility in children with CL/P. The results of Kruskal–Wallis test indicated a statistically significant difference between the 4 groups with different rates of hypernasality (normal resonance, mild, moderate, and severe hypernasality) in

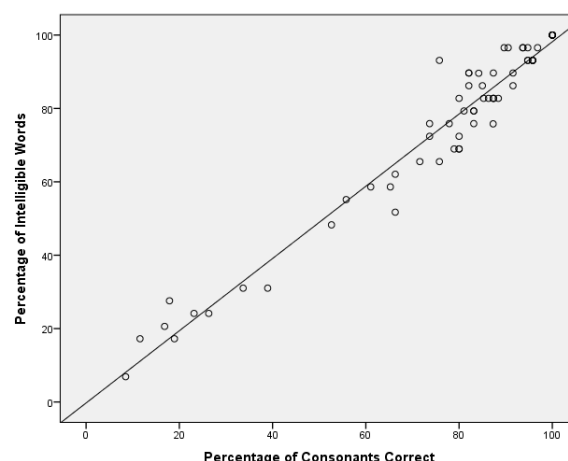


Fig. 1. The Relationship between percentage consonants correct (PCC) and percentage intelligible words (PIW) in children with CL/P

PCC ($p = 0.001$) and PIW ($p = 0.001$).

The results of the present study revealed that all children with CL/P had compensatory and obligatory errors. In this study, 19 participants (59.4%) out of 32 had compensatory errors and 13 (40.6%) had obligatory errors. The results of Mann–Whitney U test indicated no significant difference between gender and PCC and PIW ($p = 0.92$).

Discussion

The primary aim of the current study was to examine the speech intelligibility of children with repaired CL/P who had not been referred for speech therapy. One of the measures used in this research was PIW which determines speech intelligibility. Based on the results of the present study, the mean PIW in Persian speaking children with CL/P was lower than the control group and lower than the mean PIW in 3 to 4-year-old normal Persian speaking children, reported by Valizade et al. (3). They reported that the mean PIW in 3- and 4-year-old normal children was 83% to 96% (3). However, the mean PIW score in 3- to 5-year-old children with repaired CL/P was 59.68%. It can be concluded that children with CL/P had lower speech intelligibility than their normal peers. In other words, the listeners could understand on average <60% of the words produced by the children with CL/P. According to Paulson et al., speech intelligibility with a range of 50-79% is considered moderately intelligible

Table 1. The percent of children by different type of CL/P (n= 32)

Frequency	Uni-lateral Cleft Lip and Palate	Bi-lateral Cleft Lip and Palate	Cleft Palate	Submucosal Cleft	Total
N	6	14	5	7	32
%	18.8	43.8	15.6	21.8	100

Table 2. Descriptive Analysis of Variables

Variable	Children With Clefts (n = 32)	Control Group (n = 32)
Age	50.2±8.5	51.0±8.3
PCC	61.77±27.58	90.35±8.61
PIW	59.68±26.61	89.54±10.41

PCC: Percent Consonants Correct, PIW: Percent Intelligible Words

(24). Thus, the speech of children who participated in this study was moderately intelligible. Gordon-Brannan and Hodson reported that the mean percentage of speech intelligibility in 4-year-old children was 93% (25). Also, Baudonck et al. estimated that speech intelligibility in children aged 4.6 to 5 years was 90% (26). According to Gordon-Brannan and Hodson, if speech intelligibility of a child older than 4 years old is less than 66%, he/she must be referred for an appropriate treatment (25). As a result, it seems that all of our participants should be referred for speech therapy services.

Speech errors have a negative effect on speech intelligibility of children with CL/P. After analyzing the relationship between PIW and PCC, we realized that PCC was increased by an increasing in PIW. As was expected, those children who could produce more words with correct consonants had higher speech intelligibility. Other studies have also been conducted in this field. For instance, Zajac et al., found a moderate correlation between speech intelligibility and PCC (27). Similarly, Willadsen and Poulsen reported a significant relationship between PCC and intelligibility scores (28).

Consistent with the previous studies, no significant difference was found between girls and boys in speech intelligibility. Van Lierde et al. studied speech intelligibility of 43 children with unilateral cleft lip and palate and reported no notable difference in the scores of intelligibility between males and females (29). Furthermore, Dames et al. studied speech intelligibility in 72 children with unilateral and bilateral cleft lip and palate and reported no significant difference between girls and boys with unilateral cleft (30). Our study group consisted of 4 different types of cleft; thus, the number of males and females was not enough for analyzing the gender effect on speech intelligibility in different types of cleft.

In the current study, 32 children with CL/P participated; of them, 59.4% had compensatory error and 40.6% had obligatory error. However, Hardin-Jones and Jones stated that from the 212 preschoolers studied, 37% had obligatory error and 25% had compensatory error (31). This difference is possibly due to the fact that our study group had not received early intervention from speech therapy services, but the participants in Hardin-Jones and Jones' study had received speech therapy (31). It can be concluded that speech therapy could decrease compensatory errors in children with CL/P because these speech sound errors are not caused by structural problems. In other words, children learn compensatory errors developmentally (13), thus, this phenomenon could reflect the difference between our study and the survey conducted by Hardin-Jones and Jones (31). Compensatory errors need to be treated by speech therapy considering the fact that children with CL/P use abnormal articulation placement that can adversely affect their speech intelligibility (32). Hence, speech therapy is essential for children with CL/P, especially those who use compensatory errors.

Normastura et al. reported a high prevalence of hypernasality and speech disorders in patients with CLP (33). Similar to McWilliams who explored a direct relationship between speech intelligibility and severity of nasality and

articulation errors (10), our findings showed that most of children with CL/P had hypernasality, ranging from mild to severe, and also indicated a relationship between different rates of hypernasality and speech intelligibility.

Hearing problems have been reported in previous literature in the children with CL/P (14, 15). Similarly, the results of the present study revealed that our participants had conductive hearing loss. Many investigations such as the study conducted by Balbani and Montovani, (34) have demonstrated the importance of hearing in development of normal articulation (35, 36). Likewise, Schönweiler et al. reported that children with conductive hearing loss had severe language problem (15). Therefore, hearing loss may lead to decreased speech intelligibility in children with CL/P.

Considering the high prevalence of structural defects in the oral cavities of children with CL/P, it has been suggested that a multidisciplinary cleft team deliver a variety of services to these children (37). The speech and language pathologist and the audiologist are members of the interdisciplinary program for clients with CL/P and their services are required for improving the communication skills in this population. As Edmondson and Reinhartsen pointed out, young children with CL/P need early intervention in the first 3 years of life (38). Unfortunately, in our study, the participants had not been referred for early intervention in speech and language pathology. It has been reported in the previous investigation that children with CL/P would have speech disorders following primary cleft palate surgery (39). Thus, it is recommended that our study group be referred to speech and language therapy following surgery.

Limitation

The first limitation of our study was the small size of the Persian speaking children with repaired CL/P aged 3 to 5 years who were not referred for early intervention in speech therapy. This small size was not enough to have robust results and conclusions. The second limitation was the diversity of cleft types in our study. Because of the number of children in each type of cleft, we could not examine the gender effect on speech intelligibility. Moreover, the third limitation was that all the children were operated on by 1 surgeon in Mashhad. The fourth limitation was that we could not have a control group with conductive hearing loss. Thus, it is suggested that the speech intelligibility of children with cleft lip and palate be compared with children experiencing conductive hearing loss in future studies.

Conclusion

The findings of the present study revealed that 3- to 5-year-old children with repaired CL/P who had not been referred for early speech therapy intervention had moderate speech intelligibility. The results of intelligibility analysis reflected that more than half of their speech was unintelligible to the listeners. Although all our participants were diagnosed with an articulation disorder affecting their speech intelligibility, none of them had any experience with early intervention for improving their

speech intelligibility following the surgery. Therefore, it is recommended that these children receive speech and language therapy services after surgery.

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Conflict of Interests

The authors declare that they have no competing interests.

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