

Research Article



Language Screening in Toddlers with Cleft Lips and or Palates: A Pilot Study

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ABSTRACT

Introduction: Children with cleft lips and or palates are at risk for speech and language disorders. Early speech and language intervention help them to advance their full speech and language abilities. This study aimed to determine whether language abilities in toddlers with oral clefts would differ from those of normal children.

Materials and Methods: The study recruited 2 groups of children aged 12 to 24 months. The toddlers with cleft lips and or palates as a group of children with orofacial anomalies were born at Shahid Akbar-Abadi Hospital from March 2017 to March 2019 in Tehran City, Iran. The parents of non cleft children and those with oral clefts answered the questions of the verbal and non-verbal communication screening checklist for Persian-speaking children.

Results: The results showed significant differences between the scores of the receptive-expressive language achieved by toddlers with clefts and non-clefts peers aged 12-18 months ($P < 0.05$) and 18-24 months ($P < 0.05$). But the results showed no significant differences between the non cleft toddlers and those with clefts in non-verbal communication ($P > 0.05$).

Conclusion: The toddlers with orofacial anomalies need early speech and language intervention.

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1. Introduction

The children produce their first words by one year of age [1, 2], and around 18 and 24 months begin to put two words, and then three words together lead to short sentences stage [3]. Receptive and expressive language skills enable children to communicate and convey their needs, feelings, and thoughts. A child with language delay or disorder may have problems in receptive or expressive language or both. Based on reports, 16% of children show a delay in the initial stages of language acquisition; probably some of them should be referred to as clinical disorders [4]. Sunderajan and Kanhere investigated speech and language delay and its risk factors. They reported that the prevalence of speech and language delay in children who attended the pediatric outpatient department of a hospital was 2.53%. They concluded that physical (oropharyngeal) deformity is one of the risk factors associated with speech-language delay [5]. One of the most frequent congenital anomalies is orofacial clefts. In Asia, the prevalence of syndromic and nonsyndromic cleft lip with or without cleft palate is reported 1.30-1.47 per 1000 live births [6]. In Iran, the prevalence of oral cleft was estimated at 1.24 in 1000 live births [7]. Children with oral clefts are at risk for communication disorders; thus, early speech and language intervention is often recommended [8-10]. Many investigations are about cleft palate speech productions, but few researchers were interested in studying the language skills of toddlers or children with cleft palate. The evidence demonstrated a language delay in children with oral clefts.

Some reports mention differences in early vocalization in the prelinguistic stage between non-clefts toddlers and those with cleft anomalies [11, 12]. Scherer et al. concluded that children with clefts use non-verbal communicative acts when verbal development is delayed [13]. Kapp-Simon KA, Krueckeberg concluded that some children with cleft lip and palate might be at risk for developmental problems in the first two years of life [14]. Speltz et al. reported that the children with clefts showed deficits in cognitive and psychomotor development. They found that the performance of cleft group infants in verbal and nonverbal areas was significantly lower than the non-cleft children [15]. There is evidence that the children with Cleft Lip and Palate (CLP) show a delay in the production of the first words [16, 17] and development of vocabulary and two-word utterances [18, 19]. Bzoch found that children with CLP have delays in expressive language at the age of 12, 18, 24, and 36 months, but their receptive language is normal

[20]. Long and Dalston studied the receptive language of one-year-old babies with cleft lip and palate and concluded that they had no comprehension deficits [21]. But Lamônica et al. found significant differences between the children with cleft lip and palate and non cleft children in receptive and expressive language skills [22].

Evidence demonstrates that toddlers who exhibit speech and language developmental delays are at risk for persistent language difficulties [23, 24]. Morris and Ozanne concluded that children who had been identified with significantly delayed expressive language development at two years of age showed expressive language problems at 3 years, too [25].

On the contrary, some researchers concluded no language deficit in cleft palate children. Priester and Goorhuis-Brouwer examined the receptive and expressive language of 43 toddlers with cleft palate and 32 toddlers without cleft palate. They found no significant differences between the non cleft children and those with Cleft Lip and or Palate (CL/P) in receptive and expressive language and articulation [26].

Early intervention is essential for children with oral clefts, but the children with cleft palate are usually referred to a Speech and Language Pathologist (SLP) after surgery if they show signs of speech and language disorders. Ghayoumi Anaraki et al. reported low-performance language abilities in 4- to 7-year-old Persian-speaking children with cleft lip and palate. They proposed the early language assessment and treatment for children with cleft lip and palate [27]. The current study aimed to answer whether the 12 to 24 months old Persian-speaking toddlers with CLP, compared to their non clefts peers, have deficits in receptive and expressive language.

2. Materials and Methods

This cross-sectional study was conducted on 72 12-24 months old children. They were two groups of children. The first group comprised 24 children with orofacial anomalies born at Shahid Akbarabadi Hospital from March 2017 to March 2019. The live births children with cleft lip and palate at the age of 12 to 24 months registered according to the code in the International Classification of Diseases (ICD 10th revision) at Shahid Akbarabadi hospital were included in this study. We gathered information from hospital medical records. The cleft lip and palate children with other problems such as hydrocephalus were excluded.

Table 1. Demographic data of the participants

Variables	No.(%)		
	Clefts	Non-clefts	
Age	12-18 mo	10(41.7)	21(43.8)
	18-24 mo	14(58.3)	27(56.3)
Gender	Male	14(58.3)	30(62.5)
	Female	10(41.7)	18(37.5)
Total	24(100)	48(100)	

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The parents of children with CLP who signed the consent form answered the questions of the verbal and non-verbal communication screening checklist for Persian-speaking children aged 12-24 months. The verbal and non-verbal communication screening checklist was developed by Safariyan et al. and reported its validity and reliability. Its Intra-Class Correlation Coefficient (ICC) was 0.80, and the Cronbach α for the total scores was ≥ 0.70 . The checklist is divided into 3 parts: receptive language, expressive language, and non-verbal communication. The checklist has yes/no questions that the parents or caregivers should answer them. One score is for "Yes" and 0 score for "No" [28].

The typically developing non cleft toddlers as the second group were randomly recruited from kindergartens in Tehran. They were 48 children whose scores on all developmental domains on the Age and Stage Questionnaire (ASQ) [29] were within normal limits. The parents of normal children who signed the consent form answered the questions of the verbal and non-verbal communication screening checklist for Persian-speaking children too.

Statistical analysis

The obtained data were analyzed using SPSS software version 21. The descriptive statistics and non-parametric testing (the Mann-Whitney U test) were conducted. Statistical significance level was set at a P-value less than 0.05.

3. Results

There were 16844 registered live births at Shahid Akbarabadi Hospital from 2017 to 2019. During the study period, 24 infants with Cleft Lip (CL) with or without Cleft Palate (CP) were born alive in the hospital. Overall, the prevalence of oral clefts was 1.4 per 1000 live births. Table 1 presents demographic data of participants divided into two groups (The children with oral clefts and non-cleft children aged 12-18 months and 18-24 months old). Table 2 presents the distribution of children with the type of clefts. Overall, of 24 toddlers with cleft lip and or cleft palate (CL \pm P), 14 (58.3%) were male. Of 24 toddlers, 41.7% had CLP, 25% CP, and 33.3% CL. All toddlers with oral clefts were treated.

Table 3 presents the descriptive and analytical statistics of total scores of each part of the verbal and non-verbal communication screening checklist in 2 age groups.

Table 2. Distribution of type of clefts based on gender

Gender	No.(%)			
	Cleft Lip	Cleft Palate	Cleft Lip and Palate	Total
Male	4(16.7)	6(25)	4(16.7)	14(58.3)
Female	4(16.7)	0(0)	6(25)	10(41.7)
Total	8(33.3)	6(25)	10(41.7)	24(100)

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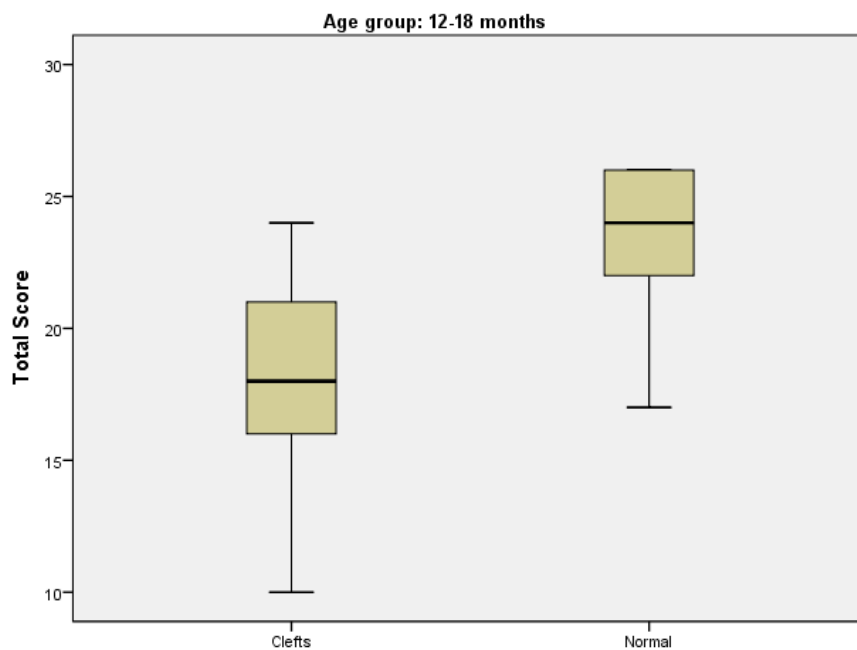


Figure 1. Box plot of total scores of two study groups at the age of 12-18 months

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The Kolmogorov-Smirnov test at $P > 0.05$ was conducted, indicating the normally distributed data; hence, a Mann-Whitney U test was used to compare the mean scores. The results showed a significant difference between the total scores achieved by toddlers with clefts and non-clefts children aged 12-18 months ($P = 0.0001$) and aged 18-24 months ($P = 0.012$). Figures 1 and 2 show the box plots that describe the total scores of the verbal and non-verbal communication screening checklist in 2 groups. The mean scores of the expressive and recep-

tive language and non-verbal communication were calculated separately in each group. The results showed a significant difference between the scores of the receptive language that toddlers with clefts and non cleft peers achieved at 12-18

months ($P = 0.003$) and 18-24 months ($P = 0.044$). The scores of the expressive languages in toddlers with clefts and non-clefts peers aged 12-18 months were significantly different ($P = 0.0001$). The expressive language scores

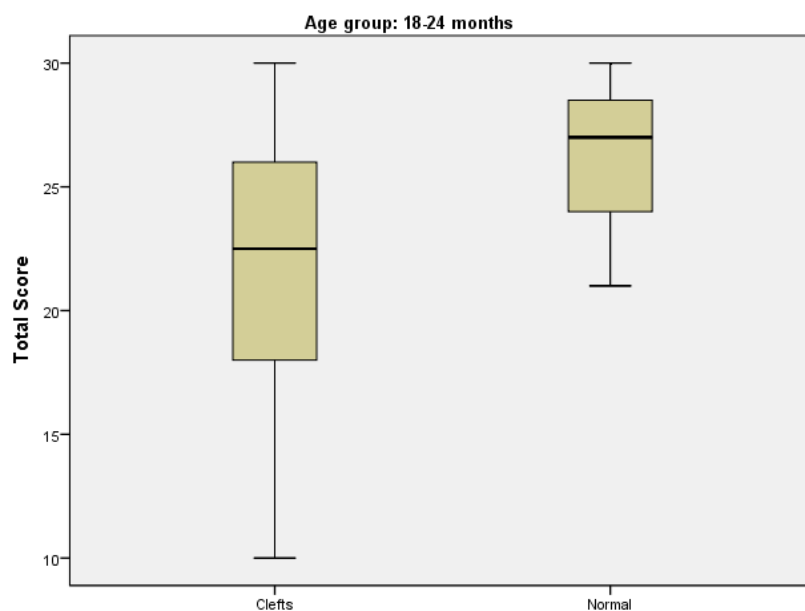


Figure 2. Box plot of total scores of two study groups at the age of 18-24 months

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Table 3. The descriptive and analytical statistics of scores of the verbal and non-verbal communication screening checklist

Children	Variables	Clefts			Normal			P-Value
		Mean±SD	Min	Max	Mean±SD	Min	Max	
12-18 months	Receptive language	4.10±1.28	1	5	4.95±0.21	4	5	0.003*
	Expressive language	5.30±3.46	2	11	9.95±1.24	6	11	0.0001*
	Non-verbal communication	8.10±2.02	5	10	8.81±1.4	6	10	0.46
	Total score	17.50±4.69	10	24	23.57±2.46	17	26	0.0001*
18-24 months	Receptive language	6.21±2.15	0	8	7.37±0.68	6	8	0.044*
	Expressive language	6.43±4.01	0	12	9.7±2.31	5	12	0.007*
	Non-verbal communication	9.36±1.15	6	10	9.44±0.97	6	10	0.88
	Total score	22.50±5.79	10	30	26.44±2.88	21	30	0.012*

* P<0.05

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in toddlers with clefts and non cleft children aged 18-24 months were significantly different, too (P=0.007). The non-verbal communication scores were not significantly different between the two groups children aged 12-18 months (P=0.46) and 18-24 months (P=0.88).

4. Discussion

The care of children with orofacial clefts requires a multidisciplinary team [30]. In many literature research and documents, the emphasis is on early intervention by speech and language pathologists for children with craniofacial anomalies [30-32]. Witzel reported that 75% of children with a repaired cleft palate require speech and language therapy throughout childhood and adolescence [33]. Some reports show that expressive language development in children with oral clefts is slower [20, 21].

We aimed to survey expressive and receptive language and non-verbal communication skills of toddlers with orofacial anomalies. The current study results revealed receptive language and expressive language delay in toddlers with CLP aged 12-24 months, but they had no problems in non-verbal communication. Many authors reported expressive language delay in children with clefts [5, 11-13]. An early speech and language intervention are recommended for training the parents of toddlers with cleft anomalies to help improve child language [13, 30, 34]. Usually, cleft lip repair is done when a baby is 3 to 6 months, and cleft palate surgery is usually done approximately at 1 year. However, some parents were not referred to Speech and Language Pathologists (SLP); thus, these toddlers lacked any early speech and language interven-

tion experiences. According to Nagarajan et al., a cleft palate child may either not be referred to SLP or be referred when the child's language disabilities are obvious [9].

Speech and language intervention during preschool age plays an essential role in improving the language skills in children [33]. The parent-child communication and having a linguistic environment at home are very important in early language development [35]. Pushpavathi et al. suggested that mothers' involvement in early language intervention helps in the language development of children with repaired cleft lip and palate. They concluded that home training and stimulation by parents significantly improve the speech and language of children with CLP [36]. Some investigators reported that children with cleft lip and palate show poor cognitive-linguistic and learning abilities than normal children [37]. The current study results show poor language ability in toddlers with CLP than normal children. Therefore, these children need early language intervention that may help relieve other problems related to language deficits like learning disabilities.

5. Conclusion

The current study indicates that toddlers with CLP are at risk for receptive-expressive language impairment. Thus, children with orofacial anomalies should be referred to an SLP for early speech and language intervention.

Ethical Considerations

Compliance with ethical guidelines

This study was approved by the Research Ethics Committee according to the ethical principles and the national norms and standards for conducting medical research in Iran (ID: IR.IUMS.REC.1398.487).

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Authors' contributions

All authors equally contributed to preparing this article.

Conflict of interest

The authors declared no conflict of interest.

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