

Research Paper: Language Sample Analysis in Children With Cleft Lip and Palate



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ABSTRACT

Objectives: Cleft Palate (CP) with or without Cleft Lip (CL/P) are the most common craniofacial birth defects. Cleft Lip and Palate (CLP) can affect children's communication skills. The present study aimed to evaluate language production skills concerning morphology and syntax (morphosyntactic) in children with CLP.

Methods: In the current cross-sectional study, 58 Persian-speaking children (28 children with CLP & 30 children without craniofacial anomalies=non-clefts) participated. Gathering the language samples of the children was conducted using the picture description method. The 50 consecutive intelligible utterances of children were analyzed by the Persian Developmental Sentence Scoring (PDSS), as a clinical morphosyntactic measurement tool.

Results: The PDSS total scores of children with CLP were lower than those of the non-clefts children. A significant difference was found between the studied children with CLP and children without craniofacial anomalies in the mean value of PDSS total scores ($P=0.0001$).

Discussion: Children with CLP demonstrate a poor ability for using morphosyntactic elements. Therefore, it should be considered how children with CLP use the grammatical components.

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Highlights

- The speech skills of children with cleft palate are more prominent than their expressive language skills. Thus, speech disorders in these children were reported more than language disorders.
- The study findings suggested a significant difference between children with CLP and healthy peers in morphology and syntax.
- Statistically significant differences were found between children with CLP and healthy peers in the mean scores of grammatical categories, including verb morphology, modal and compound verbs, grammatical morphemes, pronouns, prepositions, and conjunctions.

Plain Language Summary

It was reported that children with CLP have speech and language problems. The speech problems of children with cleft palate are more distinguished than the other aspects of language skills. The current study results suggested that children with CLP cannot use grammatical items in their sentences, as much as non-clefts children.

1. Introduction

Among the most common birth defects are Cleft Lip (CL) and Cleft Palate (CP) [1]. A large body of literature reported the prevalence of clefts of lip and palate. Panamonta et al. conducted a systematic review to assess the available evidence concerning the birth prevalence of orofacial clefts, including CL, Cleft Lip and Palate (CLP), and CP. They reported that among races, the prevalence of oral clefts ranged between 2.62 and 0.58 per 1000 live births [1].

Khazaei et al. concluded that the CP with or without cleft lip (CL/P) incidence in Iran was equal to 1/1000, i.e., a lower incidence, compared to other countries [2]. Haseli et al. reported that the overall prevalence of CL and CP in Iran was lower than those of other countries in the Middle East and Asia [3].

Researchers have long proved that children with CLP are at risk for speech and language disorders. Schönweiler et al. reported that 92% of children with CP presented speech and language disorders [4]. Some researchers signified language delay and disorders in various-aged toddlers and children with CLP. Jocelyn et al. compared receptive and expressive abilities between children with CLP and non-cleft peers at the age of 12 and 24 months. Accordingly, they concluded that children with CLP significantly obtained lower scores on expressive language abilities, compared to the controls [5].

Scherer reported that toddlers with CLP encountered a delay in expressive language [6]. Fox et al. examined language in children with CLP and CP, aged 24-33 months. Subsequently, they found that the expressive language of the CP group was different from that of the control group [7]. Scherer et al. studied speech and language milestones in children with CP, aged 6-24 months. They collected the speech samples of children at their home and analyzed 100 utterances of them. Scherer et al. documented that some children with CP failed to produce first words until the age of 18 months.; at 24 months of age, they did not use multi-word utterances [8]. Lamônica et al. concluded that children (aged between 12 & 36 months) with CLP exhibited poorer performance in receptive and expressive language, compared to those children without such conditions [9]. Richman and Eliason reported language disabilities in school-aged children with CP [10].

Prudenciatti et al. compared the cognitive-linguistics skills of children with CLP and children without craniofacial anomalies, aged between 5 years and 6 years and 11 months. They concluded that the language skills of children with CLP required for academic tasks were lower than their healthy counterparts [11].

Ghayoumi Anaraki et al. evaluated language performance in Persian-speaking children with CLP, aged 4-7 years using the test of language development. The related results indicated a significant difference between language abilities in children with clefts and normative data [12]. Peterson-Falzon et al. mentioned that children with clefts experience significant problems in the verbal output. According to Peterson-Falzon et al., all children

with clefts should be considered for language development. They proposed the Developmental Sentence Scoring (DSS) to measure sentence complexity [13].

The current study aimed to evaluate expressive language in Persian-speaking children with CLP. We mainly addressed how Persian-speaking children with CLP can use grammatical components in their sentences.

We used a clinical measurement tool, called the Persian Developmental Sentence Scoring (PDSS) to assess the morpho-syntactic abilities of language sample among Persian-speaking children [14]. The psychometrics information of the PDSS, including inter-rater reliability (0.77), internal consistency (0.79), convergent validity (0.97), and age discriminative validity (0.69) revealed that it can be used as a reliable numerical measurement to analyze the Persian grammatical categories.

The mean PDSS total scores and the mean scores of Persian grammatical categories were compared between two study groups; children with CP or CLP and children without craniofacial anomalies (non-clefts). The PDSS grammatical categories include the following aspects: verbal morphology, modal and compound verbs, grammatical morphemes, pronouns, question words, prepositions, and conjunctions.

2. Methods

In total, 58 monolingual Persian-speaking children participated in this study. The study participants included two groups; a group consists of 28 children with non-syndromic CP or CLP aged from 4 to 6 years; an age- and gender-matched comparison group, including 30 Typically-Developing (TD) children without craniofacial anomalies. Furthermore, children with CP or CLP were recruited from rehabilitation centers (speech therapy clinics) in Tehran City, Iran. None of the children exhibited other congenital anomalies, sensorineural hearing loss, and neurological impairments (the children who had unintelligible speech & those with syndromic CLP were excluded), and TD children were recruited from kindergartens in Tehran City, Iran. TD children presented no history of language delay, neurological impairments, and congenital anomalies. Their parents completed the Age and Stage Questionnaire (ASQ) [15]. The Persian version of the ASQII is used for screening developmental disorders in children. The reliability of the ASQ was reported to range from 0.76 to 0.86; the inter-rater reliability of the scale was calculated as 0.93.

Their scores of the developmental domains (communication, fine motor, gross motor, personal, social, & problem-solving skills) were within healthy limits.

This study was approved by the Ethics Committee of the Iran University of Medical Sciences (Code: IR.IUMS.REC.1397.444). The informed consent form was signed by the studied children's parents. They were allowed to discontinue participation in the study at any time. Gathering the language sample of children was performed via picture description. The examiner sat in an appropriate room (with minimum noise & enough light) with each child and requested them to explain the pictures that illustrated daily activities at home, the physician's office, a birthday party, and the seaside. During conversations, a Kingston-DVR-902 digital voice recorder was used. The examiner, as a Speech and Language Pathologist (SLP), evaluated 5 components, including facial grimace, nasality, nasal emissions, phonation, and articulation based on the Pittsburgh Weighted Speech Scores (PWSS). The PWSS is a quantitative clinical scale for auditory perceptual assessment. A score of ≥ 7 indicates Velopharyngeal Insufficiency/Incompetence (VPI) [16].

The language samples of children were orthographically transcribed by the examiner. Subsequently, the utterances with the following specifications were excluded: one-word utterances, utterances without a verb, and unintelligible utterances. The language samples consisted of a maximum of 50 consecutive intelligible utterances. The scores of each grammatical category used per utterances (sentences) were calculated. Then, the following formula was employed: total grammatical scores divided by 50=PDSS for the language sample of each research participant [17].

Concerning the reliability estimates for transcription, the language samples of 12 (20%) study participants were randomly selected. Besides, a second transcriber transcribed them again. One of the percentages of agreement, i.e., the point-to-point percentage of agreement was conducted as a measure of interrater agreement. The formula used for percentage agreement calculation was as follows: total agreement/total agreement+total disagreement $\times 100$ [17]. The agreements and disagreements were calculated for the units of utterances (morphemes). The collected results revealed 89.1% inter-rater agreements for morphemes.

Respecting the reliability estimates for the PDSS scores, the PDSS of Language samples of 12 (20%) study participants were re-calculated to examine the inter-rater

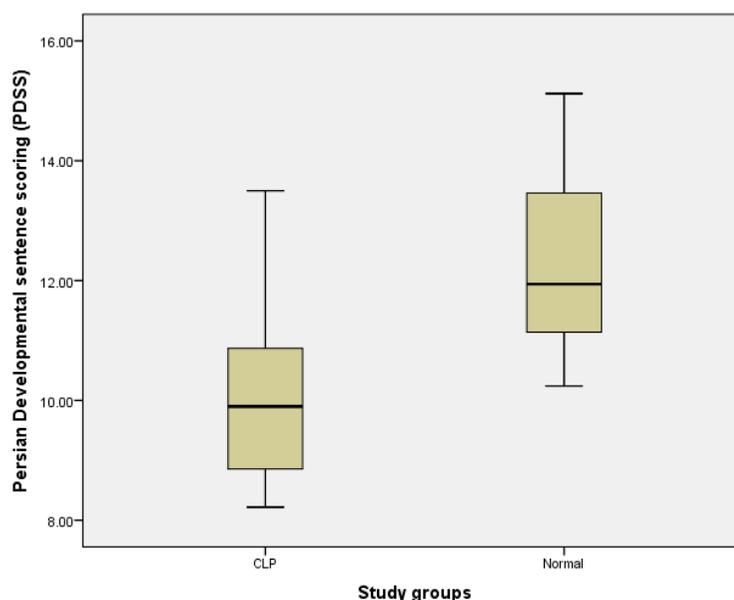


Figure 1. The mean PDSS scores of the study groups

reliability [17]. The related findings indicated a 90.1% inter-rater agreement for the PDSS scores.

All Statistical analyses were performed in SPSS v. 21. A Kolmogorov-Smirnov normality test was conducted to examine the normal distribution of the obtained data at $P > 0.05$. The non-parametric Mann-Whitney U test was also applied. $P < 0.05$ was considered statistically significant.

3. Results

The current study examined 58 children (26 girls & 32 boys). Table 1 presents the demographic characteristics of the study groups. Table 2 lists the speech-related characteristics of children with non-syndromic CLP. Based on perceptual analysis data, most of the studied children (92%) were diagnosed with mild to severe hypernasality and the presence of nasal air emission. The compensatory articulation errors (glottal stop, pharyngeal fricative, or pharyngeal stop) were elicited from the speech samples

of 14 (50%) of the explored children with CLP. Four children manifested voice problems.

Table 3 and Figure 1 demonstrate the descriptive statistics of PDSS scores of the research groups. A significant difference was found in the mean PDSS scores ($P = 0.0001$) between the investigated children with CLP and their TD counterparts. Table 3 lists the Mean±SD total scores of each grammatical category in children with CLP and those without craniofacial anomalies. There was a significant difference between the studied children with CLP and the non-cleft group concerning verb morphology ($P = 0.001$), grammatical morphemes ($P = 0.0001$), pronouns ($P = 0.015$), question words ($P = 0.006$), prepositions and conjunctions ($P = 0.001$), sentence type ($P = 0.047$), and correct sentence scores ($P = 0.024$).

4. Discussion

Disorders in the components of speech, including resonance and articulation in children with CLP, are recog-

Table 1. The demographic characteristics of the study participants

Groups	N	Age (y)		Gender		Cleft Types	
		4-5	5-6	Male	Female	CLP	CP
CLP	28	14	14	15	13	22	6
TD	30	15	15	17	13	-	-
Total	58	29	29	32	26	22	6

Table 2. The speech-related characteristics of the explored children with non-syndromic clefts

Participants	Compensatory	Hypernasality	Voice Problem
	Articulation Errors	Nasal Emission	
1	-	-	-
2	+	+	+
3	-	+	-
4	-	+	-
5	+	+	-
6	-	+	-
7	+	+	-
8	+	+	+
9	-	+	-
10	+	+	-
11	+	+	-
12	+	+	-
13	-	+	-
14	+	+	-
15	-	-	-
16	+	+	-
17	+	+	-
18	+	+	+
19	-	+	-
20	+	+	-
21	-	+	-
22	-	+	+
23	-	+	-
24	+	+	-
25	+	+	-
26	-	+	-
27	-	+	-
28	-	+	-

Table 3. The descriptive statistics of the PDSS and grammatical categories scores between the studied children with CLP and TD children

Measures	Groups	Mean±SD	P
PDSS	CLP	10.09±1.38	0.0001
	TD	12.31±1.46	
Verb morphology	CLP	91.00±13.48	0.0001
	TD	108.16±10.53	
Modal and compound verbs	CLP	29.39±9.73	0.082
	TD	36.86±15.59	
Grammatical morphemes	CLP	168.89±24.03	0.0001
	TD	212.33±30.6	
Pronouns	CLP	20.25±13.42	0.015
	TD	31.33±18.08	
Question words	CLP	0.14±0.59	0.006
	TD	11.53±2.67	
Prepositions & conjunctions	CLP	49.07±27.33	0.0001
	TD	79.66±22.75	
Sentence type	CLP	50.00±0.001	0.047
	TD	50.26±0.82	
Correct sentence	CLP	49.10±1.34	0.024
	TD	49.43±1.94	

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nizable and prominent. Consequently, the goals of intervention will be focused on speech therapy. According to some scholars, expressive language in children with CLP was poorer than their TD peers [5-11]. The present study explored the expressive language ability of children with CLP. The PDSS is a clinical measurement to assess the language sample of Persian-speaking children [14]. The PDSS was used for studying the morpho-syntactic abilities of children with CLP. The obtained findings indicated a significant difference in the dimensions of morphology and syntax between children with CLP and TD children without craniofacial anomalies. The collected data suggested that the mean scores of the PDSS and the mean scores of grammatical categories were significantly different between the investigated children with CLP and their TD counterparts. As a result, the studied children with CLP presented the ability to use sentences for describing pictures; however, their abilities concerning

morphology and syntax were below the TD children. Furthermore, the studied children with CLP used sentences with lower scores than those applied by the non-clefts children. In other words, the ability of the children with CLP for using morphosyntactic elements was lower than that of their TD peers. The current study results were in line with those of the previous findings, highlighting that Persian-speaking children with CLP face difficulties in acquiring grammatical categories [10]. The collected results supported the findings of Lamônica et al. as well as Prudenciatti and associates [9, 11].

Previous studies concluded that language deficits are related to phonological disorders [18], compensatory articulation [19], parent interaction, and stimulation [20]. Thus, speech and language pathologists should evaluate speech and language development in children with cra-

niofacial anomalies; accordingly, they should consider the aspects of speech and language for treating these children.

5. Conclusion

The obtained evidence revealed that the explored children with CLP presented lower expressive language ability, compared to their TD peers. Therefore, language abilities in children with CLP should be evaluated and considered for interventions accompanied by speech therapy.

Ethical Considerations

Compliance with ethical guidelines

This study was approved by the Ethics Committee of the Iran University of Medical Sciences (Code: IR.IUMS.REC.1397.444). The informed consent form was signed by the studied children's parents. They were free to leave the study whenever they wished.

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Authors' contributions

Methodology, Investigation, Writing – original draft, and Writing – review & editing: Leila Safarpour, Ali Ghorbani, and Nahid Jalilevand; Conceptualization: Ali Ghorbani, Nahid Jalilevand; Methodology & Data collection and analysis: Leila Safarpour, Mahboobeh Rasouli, Golamreza Bayaziyan.

Conflict of interest

The authors declared no conflicts of interest.

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