Research Paper: The Effects of Intensive Speech Therapy on Non-oral Cleft Speech Characteristics and Quality of Life in Children With Cleft Palate

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Citation: Baghban K, Derakhshandeh F, Adibi A, Shati M, Khanlar F, Mostajeran F, Zarifian T. The Effects of Intensive Speech Therapy on Non-oral Cleft Speech Characteristics and Quality of Life in Children With Cleft Palate. Iranian Rehabilitation Journal. 2021; 19(3):315-324. http://dx.doi.org/10.32598/irj.19.3.24.5

http://dx.doi.org/10.32598/irj.19.3.24.5

Article info:
Received: 28 Apr 2021
Accepted: 04 Aug 2021
Available Online: 01 Sep 2021

Keywords:
Cleft palate, Children, Speech therapy, Quality of life, Non-Oral cleft speech characteristics

ABSTRACT

Objectives: This study aimed at investigating the effects of speech intervention on Non-Oral Cleft Speech Characteristics (NCSCs), intelligibility, and Quality of Life (QoL), as well as the relationship between speech variables and QOL-related variables in intensive speech therapy among children with cleft palate.

Methods: This single-group pre-test and post-test clinical trial study was performed on 12 Persian-speaking children with cleft palate and NCSCs aged 3-7 years. For this purpose, we used 30 sessions of speech therapy and the Persian version of the Pediatric Quality of Life Inventory (PPedsQL). The pre-test and post-test assessments included perceptual assessment using the CAPS-A and QoL assessment using the Parent Proxy of the Persian version of the PPedsQL. The Wilcoxon's test was applied to analyze group differences in the NCSCs, the intelligibility of speech, the total score of PPedsQL, physical, emotional, social, and school functions before and after providing the speech therapy.

Results: The obtained results indicated that the frequency of NCSCs, the intelligibility of speech, the total score of PPedsQL, and its subscales significantly improved after the intervention ($P<0.001$). However, physical function presented no significant change after intensive treatment ($P= 0.15$). Additionally, the frequency of NCSCs had a significant negative c with a total score of PPedsQL and subscales. The intelligibility of speech was significantly and positively related to the total score of PPedsQL and its subscales.

Discussion: This study demonstrated that intensive speech therapy was an effective approach in improving the intelligibility of speech and QoL in children with cleft palates. moreover, this study revealed that speech variables were significantly related to QoL variables.

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Highlights

• This study indicated that applying intensive phonological speech therapy was effective in improving speech intelligibility and NCSCs among children with cleft palate.

• Increasing the intelligibility and decreasing NCSCs of speech following intensive speech interventions improves cleft palate children's QoL.

• The current research data indicated that intensive speech therapy in children with cleft palate and NCSCs should concentrate on the early intervention of speech problems. Such measures could help to reduce the consequences of social, emotional, and school function deficits.

Plain Language Summary

Due to the close relationship between speech and the child's experiences and their perception of wellbeing, there was a need to survey QoL in children with cleft palate. This study explored QoL in children with cleft palate and non-oral cleft speech characteristics before and after speech therapy. In the present study, assessing QoL was performed to survey the output of speech therapy in children with cleft palate and NCSCs (we used the parent proxy of PPedsQL that measures the comprehensions of parents about their child's QoL). It was important to understand the effects of the outcome of treatment and clinical care on QoL in these children. The parents' judgment of children's wellbeing and QoL before and after speech therapy was used in this study.

1. Introduction

Childhood speech and language difficulties result in immediate and lasting negative effects on children's lives. There are significant correlations between speech and language problems and the social and personal experiences of children [1, 2]. These extensive zones of children's social and personal experiences concern Quality of Life (QoL) [3]. The QoL indicates an individual's understanding of their wellbeing as an essential parameter for evaluating the intervention's outputs [4]. Health-Related Quality of Life (HRQOL) is a common approach used to understand patients' health status on their performance and wellbeing [5, 6].

One of the most prevalent craniofacial anomalies is cleft lip and palate [7]. A cleft palate affects speech, swallowing, and multiple other psychological aspects of a child's life. Some studies indicated that children with cleft palate and greater speech disorders were more prone to encountering issues in self-esteem, learning, school achievement, anxiety, and depression [8]. Thus, speech disorders in children with cleft palates can severely affect their QoL [9]. Several studies suggested that the social and personal factors in children with cleft palate were affected by decreased intelligibility of speech in this population [10].

Speech disorders in cleft palate patients are articulation, voice, and resonance disorders. The Non-Oral Cleft Speech Characteristics (NCSCs) are articulation productions with abnormal placement (posterior position in the vocal path at the placements of pharyngeal & glottal); due to the individual's response to abnormal structure, or abnormal physiology [11]. Some studies revealed the speech problems in children with cleft palate are imputed to the anatomical defects caused by cleft palate and cognitive-linguistic characteristics [12, 13].

Furthermore, studies suggested that these NCSCs have a phonological origin with a phonetic exterior appearance [14]. Approximately 40% of these children experience speech disorders related to the cleft palate; their disorders remain stable throughout life [15]. Phonetic approaches are usual in the correction of NCSCs in patients with cleft palate. However, phonological approaches believe that articulation should be targeted as an indispensable part of sound speech development in any intervention [16]. Studies comparing the time duration of NCSCs correction in children with cleft palate using phonetic, phonological, and naturalistic interventions revealed that therapies that result in phonological approaches are obtained in a shorter time [17, 18].

The relationship between speech and the child's experiences of wellbeing (QoL) is critical. A cost-effective study has indicated that excessive healthcare expenses

are expended on short-term and long-term health (e.g., speech) and social and economic development in populations with speech disorders [19]. However, there is no study with valid tools to indicate whether speech therapy has significant positive effects on the QOL of children with cleft palate. Additionally, there is no evidence concerning the effects of intensive speech therapy on QoL in children with cleft palate; therefore, our information remains inadequate in this respect.

The only research available in Persian on QoL in individuals with cleft palate was conducted by Jafari Rasa and associates (2021). In their study, the psychometric properties of the QoL adolescent cleft (QoLAdoCleft) questionnaire were reviewed [20]; however, they overlooked the correlation between QoL and speech. Accordingly, we aimed to measure QoL before and after the intensive speech therapy (2 months) in Persian-speaking children with cleft palate. It is essential to recognize the impact of clinical care on QoL in these children; thus, this study measured the variation in QoL before and after intensive speech therapy. We also analyzed the correlation between speech changes and QoL. It is crucial to correct cleft palate speech disorders for living independently and participating fully in children with cleft palate in society. The first purpose of this study was to survey the QoL, NCSCs, and intelligibility of speech before and after intensive speech therapy in children with cleft palate. Moreover, the second purpose of the study was to determine the relationship between the NCSCs and intelligibility of speech, and QoL variables (the total score of PPedsQL, physical function, emotional function, social function, & school function) in the presence of the intensive speech intervention. In this study, we used the phonological approach, assuming that the timing and content of this approach could affect QoL in children with cleft palates. The goals of the intensive treatment depend on the active phonological rules in the child's mind. Besides, the therapy plan focuses on correcting the sounds group, i.e., similarly treated in the child [18].

Finally, our purposes were to enhance the developing literature on how speech interventions relate to every patient's QoL. Moreover, we intended to provide a better comprehension of how speech related to all indicators of QoL could be useful to know methods to make better the QoL for children with cleft palate.

2. Materials and Methods

Data collection was performed between December 2018 and May 2019 at the cleft lip and palate clinic of Isfahan University of Medical Sciences in Isfahan City, Iran. The statistical sample consisted of 12 children who were selected by convenience non-probability sampling method. All study participants with cleft palate were Persian-speaking monolingual children aged 3-7 years. They were under the care of the team (the patients underwent two surgeries; primary surgery at 3-4 months and secondary surgery at 12 months), with at least one NCSC, and normal hearing without any identified syndrome or severe cognitive impairment; it was identified based on patient's records in the Isfahan cleft lip and palate clinic. None of the examined patients received speech therapy in other centers or surgery during the speech therapy program.

The study participants were excluded if unwilling to cooperate during the study. The demographic profiles of the study participants are presented in Table 1.

Cleft Audit Protocol for Speech-Augmented (CAPS-A): The speech perceptual assessment was performed with CAPS-A. The CAPS-A is an instrumental with good psychometric properties, i.e., appropriate for use in the inter-center investigation of cleft palate speech. The mean score of criterion validity in each part was equaled to 87% (range: 70%-100%). In seven parts, intra-rater and inter-rater reliability values were estimated to be good and very good, respectively (Kappa: 0.61-1.00); these values were estimated as moderate for 3 parts (Kappa: 0.41-0.60) [11].

The CAPS-A has 8 subgroups, consisting of 1- intelligibility, 2- voice, 3- resonance, 4- nasal airflow 5- a grimace, 6- consonant production, 7- cleft type characteristics, and 8- perceived needs summery.

The subgroup a6 in the CAPS-A test was used in this study with some modifications for the NCSCs variable. We also used the percentage change method. This subgroup is consonant production; if the target consonant is correctly realized in word-initial and word-final positions circle the target phoneme in the center box; if incorrectly realized, these should be transcribed in the appropriate boxes for either word-initial or word-final positions. A 5-point scale is used for the intelligibility of speech in CAPS-A, as follows: 0= normal to 4= understanding is not possible [11].

The Persian version of the Pediatric Quality of Life Inventory (PPedsQL™ 4.0™ 4.0): The PedsQL was instructed by James Varni [5]. This 23-item scale is a self-report questionnaire with 4 subscales. These subscales assess various dimensions of QoL and include physical, emotional, social, and school functions (school & kinder-

garten functions). Each physical, emotional, and school function has 5 questions and only the physical function has 8 questions. This questioner includes two reports, as follows: the child self-reports for children between 8-18 years and the parent proxy report for children between 2-5 years. A 5-point scale is used across the questions of both reports, as follows: 0= never; 1= almost never; 2= sometimes; 3= often; 4= almost always [5, 6].

The PPedsQL is a reliable tool in pediatric health investigations. This questionnaire discriminated between healthy children and children with chronic diseases (the mean score of healthy children was 12.3 higher than that in children with chronic diseases, P<0.001). The Kappa analysis for content validity indicated 95% agreement for parent-proxy and 91% agreement for parent-proxy between two raters. The collected Cronbach's alpha coefficient values were >0.7 for children's self-reports. Furthermore, the reliability of parent proxy reports was much lower than 0.7 for 2-4 year-olds. Construct validity, content validity, and criterion validity values were suitable [6].

The NCSCs were assessed based on subgroup a6 in the CAPS test [11]. All research participants were requested to produce the speech sample in Persian. This speech sample included sentences with high-pressure consonants based on the principle of the Universal Parameters system (Table 2) [21].

The intelligibility of children's speech was judged using a short conversational speech (counting 1-10, reading poem) at the start of speech sampling. This study was a single-group pre-test and post-test clinical trial. Assessments were performed once before treatment and once after treatment. The purpose was to measure alterations in the speech variables that consist of NCSCs and the intelligibility of speech. Moreover, QoL variables consisted of the total score of PPedsQL, physical function, emotional function, social function, and school function before and after articulation therapy. Assessments included perceptual assessment using the CAPS-A [11] and QoL assessment using the parent proxy of the Persian version of the PPedsQL; this tool measures the comprehensions of parents about their child's QoL [6].

One of the researchers conducted data collection in an acoustic space in the Isfahan cleft lip and palate clinic. Initially, the perceptual assessment was exclusively conducted for each study participant before the treatment in a quiet space. After describing the procedure of assessment for the patient, they were demanded to accurately articulate speech samples, like the presented auditory pattern. Concurrently, the parent proxy of PPedsQL was completed by the child's mother. Then, the treatment for the participant was started on the next day. One day after the end of the treatment procedure, the perceptual assessment and completion of the parent proxy of PPedsQL were re-performed by the mother. For data collection aims, the assessments' and interventions' audio and video files were simultaneously recorded using a Sony AX412 IC sound recorder and a Sony NODCR/BVD 708 E camera. Data analysis was performed based on the examined patients' audio and video recordings. To listen and transcribe the recorded audio and video files, they were transferred to an MSI CX620 laptop. Next, all the audio files were encoded in the English alphabet by one of the authors and prepared for analysis in random order. The treatment sessions were conducted by one of the authors (K.B) with a 9-year of experience in cleft speech therapy.

The intensive intervention consisted of 2 months of 30 speech therapy sessions. Four 45-minute weekly speech therapy sessions were performed by the speech-language pathologist. Additionally, the study participants had daily home practice with their mother (3 times/day). In the first therapy session, the parent was given a notebook in which all the methods of the therapy sessions were described. The speech-language pathologist described the speech tasks to exercise at home and wrote them down in a notebook at the end of each therapy session. The parent participated in all therapy sessions to observe the speech-language pathologist and ask questions. Then, the speech-language pathologist observed the parent while performing these tasks. The aims of the intervention were tailored for each study participant by the speech-language pathologist. The intervention was conducted by a speech-language pathologist who specialized in the therapy of speech disorders in cleft palate using the phonological approach.

In the phonological approach, the following phenomena were pursued: the aims of intervention were determined by the phonological rules in the child's mind; the treatment plan was concentrated on the correction of sound groups that the child seemed to behave similarly. Errors were corrected at the rule level (e.g., all fricatives replaced by pharyngeal fricatives). Finally, affirmation was put on the organization of formerly natural phonological contrasts. For instance, the child who replaced all affricative consonants with stop consonants could achieve positive feedback from the therapist when every affricative was used, even though errors of articulation placement and voicing were insisted.

Two speech-language pathologists, who specialized in cleft palate speech disorders for 8 and 10 years, rated the speech samples. To measure the reliability, Intra-class Correlation Coefficient (ICC) was performed for NCSCs. To gain inter-rater agreement, half (n=12) of all speech samples in this study in addition to 12 formerly speech samples from the filing of cleft lip and palate clinic in Isfahan were randomly selected (N=24); accordingly, they reflected the set of speech products. ICC in rating NCSCs was measured as 95% and ICC in rating the intelligibility of speech was equal to 92% for the two raters. Since two speech-language pathologists had a good inter-rater agreement, the analysis was undertaken by one individual.

The statistical analyses were performed in SPSS v. 21 at a significance level of 0.05. The general characteristics of the study sample were analyzed by descriptive statistics.

The number of patients (N<30) was 12; thus, the Wilcoxon's test was executed to analyze group differences in the NCSCs, the intelligibility of speech, the total score of PPedsQL, physical, emotional, social, and school functions before and after the intervention. The Spearman rank correlation test was performed to explore the potential relationship between NCSCs, the intelligibility of speech, quality of life, physical, emotional, social, and school functions. For this purpose, we used different scores of NCSCs, the intelligibility of speech, the total score of PPedsQL, physical, emotional, social, and school functions before and after treatment.

3. Results

Twelve 3-12-year-old children with cleft palate from the Isfahan cleft lip and palate clinic were surveyed in the current research. The Mean±SD age of the study participants was 5.53±1.17 years at the beginning of the study.

To address the first purpose of the current study and assess the effect of intensive treatment on NCSCs and intelligibility of speech and QoL, the Wilcoxon's test was used. According to Table 3, NCSCs significantly decreased after intensive treatment (P<0.001) and intelligibility of speech, the total score of PPedsQL, emotional function, social function, and school function significantly increased after intensive treatment (P<0.001). However, physical function presented no significant changes after intensive treatment (P=0.15).

The second purpose of the current study was to determine the relationship between NCSCs and the intelligibility of speech with the total score of PPedsQL, physical function, emotional function, social function, and school function. Spearman rank correlation data indicated a significant

Table 1. The study participants' demographic characteristics

Demographic Characteristics	Number of Participants	Age (year)	Gender	Cleft Type	Mothers' Educational Level
Cleft palate participants	1	5.2	male	BCLP*	Graduate
	2	4.9	male	BCLP	Diploma
	3	6.3	male	BCLP	MSc
	4	5.8	male	BCLP	MSc
	5	5	male	BCLP	Graduate
	6	3.10	female	BCLP	Diploma
	7	4.6	female	BCLP	Post-diploma
	8	6.7	female	BCLP	Graduate
	9	6.9	male	BCLP	Post-diploma
	10	4.11	female	BCLP	Graduate
	11	6.5	male	BCLP	Graduate
	12	6	female	BCLP	MSc

* BCLP: Bilateral Cleft Lip and Palate.

Table 2. The list of the sentences of Universal Parameters in Persian

Target Sound	Sentences	Persian Sentences	English Sentences
/p/	Tup e puja pare shod	توپ- پویا پاره شد	Poya's ball was torn
/b/	Baba sib bede	بابا سیب بده	Dad, give me an apple
/f/	Fil ruje barf raft	فیل روی برف رفت	The elephant went on the snow
/t/	Tuti je tut did	طوطی یه توت دید	The parrot saw a berry
/d/	Hoda dar zad	هدی در زد	Hoda knocked on the door
/s/	Pesar sos dare	پسر سس داره	The boy has sauce
/z/	Reza saz zad	رضا ساز زد	Reza played a musical instrument
/ʃ/	ʃiʃ ta paʃe	شیش تا پشه	Six mosquitoes
/tʃ/	Batʃe tʃar ta pitʃ dare	بچه چارتا پیچ داره	The child has four screws
/dʒ/	dʒudʒe ye havidʒ did	جوجه یه هویج دید	The chicken saw a carrot
/k/	Je kejke kutʃik	یه کیک کوچیک	A small cake
/g/	Je galle gorg	یه گله گرگ	A herd of wolves
/q/	Kalaq qar qar kard	کلاغ قارقار کرد	The crow cawed
/x/	Xale jax xarid	خاله یخ خرید	Aunt bought ice
/m/	Ma hamum mirim	ما حموم می ریم	We go to the bathroom
/n/	Mona nun dare	مونا نون داره	Mona has bread

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negative relationship between the total score of PPedsQL, emotional functions, social functions, and NCSCs. Furthermore, a significant positive relationship was observed among the total score of PPedsQL, emotional function, and social function, and the intelligibility of speech (Table 4). Besides, a significant negative relationship was detected between the difference of NCSCs and the difference of intelligibility of speech (rs= -0.85, P<0.001).

4. Discussion

This study surveyed the effects of intensive speech therapy (30 sessions) on QoL in 12 Persian-speaking children with cleft palate. The present study data indicated that intensive speech therapy decreased the NCSCs and increased the intelligibility of speech. Moreover, the total score of PPedsQL, emotional function, social function, and school function were improved after intensive speech therapy.

The intervention program was projected in our study to enforce 4 therapy sessions every week with the phonological approach for the modification of NCSCs. Efficient and cost-effective therapy for children with cleft palate requires attention to the treatment procedures and other factors related to the success of the intervention. Furthermore, the diagnostic methods and the type and duration of intervention were evaluated in the present study. This study supported the findings presented by Derakhshandeh et al. (2016) on Persian-speaking children; they suggested that articulation therapy remarkably reduced NCSCs in them. However, they used phonetic and phonological approaches in therapy [22]. We used only phonological treatment in the correction of NCSCs. This is because a phonological approach assumes articulation as an integral part of superior levels of language structure, i.e., mean semantic, syntactic, and pragmatic sciences. Pamplona et al. (1999 & 2004) argued that the phonological approach significantly decreased the time required for the modification of NCSCs in children with cleft palate, compared with phonetic and naturalistic approaches [17, 18]. This study provided further evidence based on intensive speech therapy for a better conclusion

Table 3. A summary statistics of variables for before and after intensive therapy by Wilcoxon's test

??	Before Therapy		After Therapy		Z	Wilcoxon's Test
	Mean±SD	Median	Mean±SD	Median		
NCSCs	94.04±1.72	92.85	8.33±2.45	7.14	-3.06	0.001*
Intelligibility	10.41±3.71	0.00	97.91±2.08	100.00	-3.14	0.002*
Total PPedsQL score	69.74±1.04	68.47	85.66±1.00	86.95	-3.06	0.001*
Physical function	98.43±1.12	100.00	99.47±0.52	100.00	-1.41	0.157
Emotional function	74.16±2.52	72.50	90.83±2.74	95.00	-3.07	0.001*
Social function	50.41±3.39	47.50	92.91±2.6	95.00	-3.06	0.001*
School function	59.02±4.03	58.34	85.41±2.32	83.34	-3.07	0.001*

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NCSCs: Non-oral Cleft Speech Characteristics; SD: Standard Deviation; *Means a significant difference at P<0.001 between before and after therapy in Wilcoxon's test.

when presented by a speech-language pathologist with parent participation in daily home practice [23].

Additionally, the present study results exhibited the intelligibility of speech significantly improved after intensive therapy, i.e., consistent with the findings of some studies on phonological-based therapy. These studies stated intelligibility was highly correlated with articulation and phonological approaches improve intelligibility by expanding the child's sound system [13].

The first purpose of this study was to investigate the QoL before and after intensive speech therapy; we aimed to evaluate the outcome of intensive speech therapy in patients with cleft palate and NCSCs. This study illustrated that intensive speech therapy for 2 months significantly and positively decreased the NCSCs and increased the intelligibility of speech; in turn, it has improved the total score of PPedsQL, emotional function, social function, and school function except for physical function. The study of Damiano et al. (2015) on the mother's perspective about HRQoL in children with cleft palate using PedsQL explained speech scores were significantly related to HRQoL scores and speech concerns are important factors that affect the HRQL of children with cleft palate [24]. Therefore, our findings of the effect of intensive speech therapy on improving QoL were in line with those of Damiano and associates (2015) [24].

Our study also aimed to ascertain the relationship of NCSCs and intelligibility of speech with the total score of PPedsQL, emotional, social, and school function. Accordingly, the total score of PPedsQL, emotional, social, and school function had a significant positive relationship with the intelligibility of speech and a significant negative relationship with the NCSCs. This finding suggested that fewer NCSCs and higher intelligibility of speech were directly related to better QoL in children

Table 4. The correlations between total PPedsQL score, emotional, Social, School functions, and the NCSCs and intelligibility of speech

Variables	NCSCs		Intelligibility	
	rs	P	rs	P
Total PPedsQL score	-0.91	0.00	0.77	0.03
Emotional function	-0.80	0.02	0.68	0.03
Social function	-0.69	0.02	0.73	0.05
School function	-0.50	0.01	0.40	0.02

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NCSCs: Non-oral Cleft Speech Characteristics; PPedsQL: Persian version of the PedsQL questionnaire; rs= spearman.

with cleft palate and NCSCs. This finding was consistent with those of Bruneel et al. (2019) on the relationship between speech and QoL in individuals with cleft palate. They indicated that speech properties can enhance to a large extent the QoL [3]. Other studies revealed that speech characteristics form the attitude and judgment of the listeners about the age, wellbeing, social, and academic condition of an individual. These researchers have stated that individuals with speech problems generally appear more negative, compared to their healthy counterparts [3, 10, 25].

The emotional and social functions have significantly improved after the intervention and were significantly related to speech variables (NCSCs and intelligibility of speech). These data supported the results of some studies on children with cleft palate and speech problems. There was a significant relationship between speech and language disorders, and social, school, and personal negative experiences among children [1]. Some studies have stated that children with language delay in preschool are at risk of social, emotional, and educational difficulties in the future [26, 27]. The current study result was consistent with those of other studies that documented NCSCs and difficulty in the intelligibility of speech can lead to poor social and emotional functions in these children. Similar to other speech and language problems, decreased intelligibility in individuals with a cleft palate may harm how they are judged by their peers and community [10, 25]. Some studies outlined a significant relationship between the values of intelligibility and some of the personal and social factors. In other words, because of the speech disorder that could affect social communication, children with decreased intelligibility are more prone to have trouble making friends with their peers [10, 25].

The school function in this study significantly changed after treatment. This result was consistent with those reported by some studies reflecting that low school performance has frequently been reported in children with cleft and approximately 50% of these children have a low school achievement. There are school difficulties in children with cleft lip and palate are well known; however, the cause of these difficulties are complicated and not fully clarified [25].

The need for intensive cleft palate-related speech and language intervention was correlated to all PPedsQL subscales except for the subscale's physical function. As per other studies, the non-existing change in the subscale physical function is not surprising because it measures alternative concepts [25].

Among the factors related to Socioeconomic Status (SES), the mother's educational level was considered in this study; if a mother was confused to complete the PPedsQoL questionnaire or practice at home, she would be allowed to resolve her ambiguities. However, other issues related to SES remained unexplored.

The present study provided the primary basis for later studies on comparison of the effectiveness of phonological, natural, and traditional interventions on QoL of children with cleft palate in the form of randomized clinical trials. We hope our study will lead to more investigations on improving therapeutic approaches in speech-language pathology and QoL in children with cleft palate.

5. Conclusion

This study explained that intensive speech therapy improved QoL in children with cleft palate. Furthermore, this study demonstrated speech variables significantly related to QoL variables. The present research data have benefits for the treatment of children with cleft palate. Intensive speech therapy should concentrate on early intervention of speech problems to decrease the effect of social, emotional, and school functions deficits and improve QoL.

Ethical Considerations

Compliance with ethical guidelines

This study was approved by the Ethics Committee of the University of Social Welfare and Rehabilitation Sciences (Ethics Code: IR.USWR.REC.1398.032). Before attending the research and after selecting the subjects, the procedure and the duration of the study were fully explained to the parents. The consent form was provided by the parents of the examined children. The child entered the study after signing the consent form by the parent. Also, the child was asked verbally about his/her willingness to attend the research.

Funding

This article was extracted from a research plan and was supported by the University of Social Welfare and Rehabilitation Sciences, Tehran, Iran.

Authors' contributions

All authors equally contributed to preparing this article.

Conflict of interest

The authors declared no conflicts of interest.

Acknowledgments

We wish to thank the University of Social Welfare and Rehabilitation Sciences Research Deputy and Cleft Lip and Palate Clinic of Isfahan University of Medical Sciences.

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