Comparison of patients’ age receiving therapeutic services in a cleft care team in Isfahan

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ABSTRACT

Background: Due to numerous difficulties in patients suffering from varieties of cleft lip and palate, their therapeutic management involves interdisciplinary teamwork. This study was conducted to compare the age of commencing treatments such as speech therapy, secondary palate and alveolar bone grafting and orthodontics between those who sought treatment early and late.

Materials and Methods: In this retrospective study, 260 files of patients with cleft lip and palate based on their age at the time of admission to a cleft care team were divided into two groups: The early admission and late admission. Both groups compared based on four variables including the mean age of beginning speech therapy, palatal secondary surgery, alveolar bone grafting, and receiving orthodontics using t-test.

Results: Based on the results, among 134 patients admitted for speech therapy, the mean age of initiating speech therapy in early clients was 3.3 years, and in the late ones was 9 years. Among 47 patients with secondary surgery, the mean age in early clients was 3.88 years, and in the late clients was 15.7 years. Among 17 patients with alveolar bone grafting, the mean age in the first group was 9 years, and in the other was 16.69 years. Among 24 patients receiving orthodontic services, the mean age in early clients was 7.66 years, and in the second group was 17.05 years.

Conclusion: There was a significant difference between the age of performing secondary surgery and alveolar bone grafting and the age of beginning speech therapy and receiving orthodontic services in early references and late references to the team.

Key Words: Cleft palate, early references, secondary surgery, speech therapy

INTRODUCTION

Today, noncommunicable chronic diseases are the greatest preventable cause of death. One of the most prevalent groups of chronic diseases is orofacial diseases, in which oral cleft and palate are one the major burden of oral diseases.[1]

Cleft lip and palate is considered as the most common congenital anomalies worldwide. Cleft lip and palate is the most common congenital maxillofacial abnormality and the fourth congenital defect.[2] It is estimated that the overall global prevalence of Orofacial clefting is

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one affected individual in every 600 newborn babies. Assuming 15,000 births/h worldwide, a child is born with a cleft somewhere in the world approximately every 2 min.\cite{3} Based on another study, the prevalence of cleft lip was 3.28/10,000, and cleft lip and palate was 6.64/10,000.\cite{4} In Iran, cleft lip or palate has the high prevalence, and it is estimated 1.79/1000 live births.\cite{5}

As these children grow, they experience a lot of problems such as severe speech disorders, psychosocial problems, upper respiratory obstruction, maxillary dental abnormalities, and pre- and post-operative complications such as bleeding. It is clear that these problems cannot be resolved by individual treatment, and need a team and interdisciplinary management to provide the best services, with the right time and lower costs, and improve psychological and emotional support.\cite{6}

Furthermore, therapeutic interventions for each of these problems including speech, nutritional, dental, and hearing problems should be provided at the right age, so that they prevent further disorders through the highest therapeutic effect. Late interventions can have negative consequences, or be ineffective. For example, the suitable age for speech therapy in this group of children is 2.5–3-year-old,\cite{7,11} and with optimal treatment management 95% of children with cleft lip and cleft palate can reach normal speech ability as their peers by the age of 5–6 years.\cite{12} In contrast, delay in speech therapy has a lot of negative consequences including fixation of speech production errors and consequent resistance to treatment, inappropriate psychological, mental, and social effects on children at school age, performing unnecessary secondary surgeries to improve speech, losing opportunities for other therapeutic interventions, spending more money and time at older ages to have more sessions of speech therapy, and its negative effects on learning.\cite{12}

Dental and maxillary abnormalities in unilateral and bilateral palatal clefts and other craniofacial abnormalities are very common. These abnormalities include losing teeth, increased number of teeth, rotating of teeth, crossbite, open bite, protruded premaxilla, and Class III malocclusion which affect speech performance because of functional intervention on both tongue tip and lips. Correcting these dental problems should be carried out according to the normal teething process. Some interventions can be simultaneously performed with sudden teething like mixed dentition phase, and some should be postponed to complete growth like orthodontics and orthognathic surgeries.

Orthodontic treatments in patients with cleft palate is carried out in two stages. The first stage or early mixed dentition is at the ages of 6–9-year-old, in patients with unilateral or bilateral cleft palate suffering from severe malocclusions and cross bites should be performed before alveolar bone grafting.\cite{10}

The second stage begins before or after permanent teething, that is, after alveolar surgery at about 9-year-old, but can be delayed to 12-year-old.\cite{10} Consequences of untimely orthodontic treatment are almost similar to those of late alveolar surgeries.

Despite having several cleft palate teams across the globe, there are few studies comparing early and late treatment modalities. Vlastos et al.\cite{13} recorded therapeutic findings of 530 children who reported to the therapeutic center for craniofacial abnormalities in Athens Hospital between 1995 and 2007. They created protocols to schedule the best time of providing services to patients. In their study, the age of 4 years was considered for beginning speech therapy and reporting phonological findings of the speech. In this study, 32% of children 7–12-year-old were evaluated and subjected to orthodontic treatment. About 9.5% needed maxillofacial surgeries, and 9.5% needed bone grafts, which were scheduled for the age of 5 years in evaluation criteria.

In another study, Austin et al.\cite{14} compared individual treatments in patients who suffered from cleft palate with team managements. In this study, 253 children with clefts of lip and palate, who were born between 1998 and 2003, were studied. Of these, 86% used team services and 24% used individual services. In the group that enjoyed individual services, the number of surgeries except myringotomy was less than the other group. In other cases such as speech therapy, audiometry, dentistry services, and genetic consultation, the group that used team services received better and earlier services than the other group.

There has been no research conducted on the team services to patients with cleft lip and palate, so the importance of interdisciplinary teamwork and providing optimal therapeutic services at the right age in the management of these patients are not well recognized. Furthermore, late delivery of therapeutic services leads to failed treatment. Therefore, this
study was conducted to compare patients who came to the team at younger ages with those who came at older ages or beyond standard (international criterion) age limits for different therapeutic services according to age at commencement of speech therapy, age at secondary surgery, age at alveolar bone graft, and age at commencement of orthodontics.

MATERIALS AND METHODS

This retrospective observational analytic study was conducted on 300 files of patients with a variety of lip and palate clefts who came to Cleft Care Team at Isfahan University of Medical Sciences, Isfahan, Iran between 2010 and 2013, and received services related to the diagnosis of cleft lip or palate. At first, 300 files of patients who were between 3 months to 35-year-old were studied. Then, patients with velopharyngeal insufficiency (VPI) without a history of cleft lip or palate and children under 1-year-old were excluded (since they did not need to alveolar bone graft, speech therapy, and orthodontics). Participants were a total of 40 cases. After reviewing the files, demographics of the remaining patients (260 patients), the extent of the cleft, accompanying abnormalities, and other medical information were extracted from the files. Then, all patients were divided into four groups based on receiving services. Patients who received speech therapy were in Group A, those who received secondary surgery were in Group B, those who had alveolar grafting were in Group C, and those who received orthodontics were in Group D. Then, patients were divided into two groups of early comers and late comer based on the age of their first visit by the cleft care team. So for Group A, the criterion for distinguishing early comers from late comers was entering the team before and after the age of 4-year-old. That is, patients who came to the team before 4-year-old were early comers, and those who came after 4 were late comers. The mean age of having alveolar bone graft was measured in both groups, and compared using independent t-test.

In patients in Group D, the distinction criterion was coming to the team before and after 9-year-old. That is, those who came to the team before 9-year-old were early comers, and those who came after 9 were late comers. Likewise, mean age of having orthodontics was measured in both groups and compared using the independent t-test.

RESULTS

According to Table 1, of 260 files, 56% were boys and 44% girls. Unilateral cleft lip and palate were the most common (39.2%), and submucosal cleft was the least common (3.8%). Furthermore, 90% of submucosal clefts were observed in girls, whereas only 10% were in boys. In contrast, bilateral cleft lip and palate were more common in boys (76.6%) than in girls (23.3%).

Of 260 files, 134 patients came for speech therapy (Group A) and the remaining did not enter this department for different reasons such as not attending to speech services, not needing speech therapy or low age.

According to Table 2, of 134 patients, 75 (56%) participants were early comers (before 4-year-old), and 59 patients (44%) were late comers (after 4-year-old). The mean age at the start of speech therapy in early comers was 3.3 years, and in late comers was 9-year-old. Independent t-test showed a significant difference between the two groups ($P < 0.05$). That

| Type of cleft                        | n (%)   | Total |
|-------------------------------------|---------|-------|
|                                     | Girl    | Boy   |       |
| Unilateral cleft lip and palate     | 40 (39.2) | 62 (60.8) | 102 (39.2) |
| Bilateral cleft lip and palate      | 10 (23.3) | 33 (76.7) | 43 (16.5) |
| Cleft palate alone                  | 22 (51.2) | 21 (48.8) | 43 (16.5) |
| Cleft lip and alveolar              | 8 (57.1)  | 6 (42.9)  | 14 (5.5)  |
| Soft palate cleft                   | 25 (52.1) | 23 (47.9) | 48 (18.5) |
| Submucosal cleft                    | 9 (90)   | 1 (10)  | 10 (3.8)  |
| Total                               | 114 (44) | 146 (56) | 260 (100) |
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Table 2: Frequency and mean age of early comers and late comers at the time of receiving services

| Time of entering team type of services | Early coming |  |  | Late coming |  |  | All clients |  | P |
|---|---|---|---|---|---|---|---|---|---|
|  | Number of clients | Mean age | SD | Number of clients | Mean age | SD | Number of clients | Mean age | SD | P |
| Speech therapy | 75 | 3.37 | 0.73 | 59 | 9.09 | 5.6 | 134 | 0.001 |
| Secondary surgery | 25 | 3.88 | 1.12 | 22 | 15.70 | 6.7 | 47 | 0.001 |
| Alveolar surgery | 4 | 9 | 2.00 | 13 | 16.69 | 4.64 | 17 | 0.006 |
| Orthodontics | 6 | 7.66 | 1.36 | 18 | 17.05 | 3.5 | 24 | 0.008 |

SD: Standard deviation

is, early comers reduced the beginning age at speech therapy.

All patients who underwent surgery to remove hypernasality and improve speech were 47 people, of whom 53% were early comers, and 47% were late comers. The mean age at secondary surgery in early comers and late comers is shown in Table 2. The mean age of participants at the time of intervention in both groups was significantly different ($P < 0.05$). That is, early comers had earlier secondary surgeries.

Of 17 people who had alveolar bone graft, 23.5% were early comers, and 76.5% were late comers. The mean age of early and late comers is shown in Table 2. The difference between these two groups is significant ($P < 0.05$), which shows that early comers had earlier alveolar surgery.

Of 24 people used orthodontics, 25% were early comers and 75% were later comers. The mean age at onset of orthodontics was 7.66 and 17.05 years in the first and the second group, respectively. The difference was significant between these two groups [Table 2].

As shown in Figure 1, the age of implementing interventions (speech therapy, secondary surgery, alveolar surgery, and orthodontics) was earlier in early comers.

**DISCUSSION**

This study showed that the mean age of receiving secondary surgery, alveolar bone grafting, speech therapy, and orthodontics was significantly different in early comers and late comers. In fact, early coming leads to the timely start of therapeutic procedures, and earlier assessment of all existing problems and disorders.

Hardin-Jones and Chapman\(^{[15]}\) conducted a study to determine the effect of early intervention on speech and vocabulary development in 2.5-year-old children and found that children who received early speech therapy had a better performance than those who did not receive treatment or received it late.

In another study by Scherer et al.\(^{[6]}\) on children, the positive effect of early speech therapy on vocabulary growth, phoneme reserve, and reduced compensatory errors was shown.

With regard to the appropriate time of the secondary surgeries, several ideas have been proposed. Some have considered the best age for secondary operation immediately after certain diagnosis of VPI.\(^{[10]}\) In fact, the age at which the child begins to continuously speak, and can cooperate with the therapist is the best time to diagnose the VPI.\(^{[10]}\) The best age to perform secondary surgery is 3.5–5 years, which follows by 90% success. As the age increases, and compensatory production errors become more chronic, the probability of success of surgery reduces.\(^{[9]}\) It is noteworthy that there is the possibility of secondary surgeries in adults, but the rate of success is low. Statistics show that the success rate of secondary surgeries at late childhood or adolescence reduced to 50%.\(^{[9]}\)

In a study among 4–5 years old children for studies the effects of early secondary surgeries, observed a low rate of nasality and more intelligible speech as compared to late surgeries.\(^{[16]}\)
For the age of alveolar bone graft, there are several opinions. Some specialists suggest the time before teeth growth (15–24-month-old),\cite{16} and some consider later surgery after the growth of permanent teeth (mixed dentition).\cite{16,17} The mean age for these surgeries is considered 6–9-years-old.\cite{18} Disadvantages of late surgeries are delaying other interventions such as speech therapy, orthodontics, irregularity of teeth, face deformity, low self-confidence, speech problems secondary to dental disorders, entering food and drinks to nose through the alveolar cleft, and infection and halitosis.

Sindet-Pedersen and Enemark\cite{19} compared early bone graft surgeries with late ones, and concluded that earlier grafts before the grow of canine teeth are more successful.

On the basis of the results, the mean age at start of speech therapy was significantly different in both groups, that is, early comers received earlier and timely (according to international standards) speech therapy. (According to what was mentioned in the introduction, the best age for speech therapy is 2.5–3-year-old.) This early intervention shows the effect of early coming to the therapeutic team. In contrast, late comers delayed treatment until the age of 9 years, so they suffered from negative consequences of the late onset of treatment. This finding was in line with Austin et al.'s results. They found that earlier reference to the team warranted higher quality services.\cite{14}

In general, it seems that in addition to late coming to the team services, other reasons for late onset of speech therapy in the second group are late reference by surgeons, lack of knowledge of parents about the necessity of speech therapy after primary and secondary surgeries, involvement of parents in surgery complications such as bleeding, infection, psychological problems, and economic problems.

With regard to the age at secondary surgery, the statistical tests showed that the mean age in both early and late comers were significantly different. This figure was similar to the international standard in the first group (early comers) but was too late for the second group. This late surgery extremely reduces the success of the surgery. Reason for this late coming can be that parents fear having several surgeries on their child, and their hopelessness about improving their child’s speech because most of the secondary surgeries fail, families refrain from them. Another reason can be the unnecessary, inappropriate, lengthy speech therapy meetings that makes parents feel that secondary surgeries are not needed.

Regarding the mean age of having alveolar bone graft and the onset of orthodontics, the results showed a significant difference between early and late comers. This finding shows that these interventions occurred at the right time in the first group while they were delayed in the second group. One of the main reasons for late surgeries and orthodontics can be the high cost of dentistry services in Iran and lack of insurance coverage for such costs. Therefore, parents do not have the psychological and economic power to think of their child’s beauty, so they either postpone them or never do them. In fact, as Austin et al. showed one of the services that can affect the outcome if timely provided is dentistry services, especially orthodontics.\cite{14}

In response to the question of what factors in the therapeutic team can reduce the age of the patients and timely therapeutic interventions, it is noteworthy to mention that timely coming to the team increases families’ knowledge of the therapeutic process, and prevents unnecessary surgeries and having speech therapy sessions for structural problems, and informs parents from the suitable age for surgeries and their complications. Furthermore, team specialists are all experienced people in the field, which prevents patients from referring to other centers, and paying unnecessary costs.

Lack of a harmonic team in the past years in most parts of Iran including Isfahan has caused to be the individually treatment of many patients, so some of the services have been delayed. We hope that this study can lay the ground for other medical centers in Iran to know about the effects of teamwork approach on the treatment of patients with clefts of lip and palate, so they can provide such services as a team.

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**Conflicts of interest**

The authors of this manuscript declare that they have no conflicts of interest, real or perceived, financial or non-financial in this article.

**REFERENCES**

1. Semb G, Brattström V, Mølsted K, Prahl-Andersen B, Shaw WC. The Eurocleft study: Intercenter study of treatment
outcome in patients with complete cleft lip and palate. Part 1: Introduction and treatment experience. Cleft Palate Craniofac J 2005;42:64-8.

2. Shoae S, Ghasemian A, Mehrabani K, Naderimagham S, Delavari F, Sheidaei A, et al. Burden of oral diseases in Iran, 1990-2010: Findings from the global burden of disease study 2010. Arch Iran Med 2015;18:486-92.

3. IPDTOC Working Group. Prevalence at birth of cleft lip with or without cleft palate: Data from the international perinatal database of typical oral clefts (IPDTOC). Cleft Palate Craniofac J 2011;48:66-81.

4. Mossey P, Little J. Addressing the challenges of cleft lip and palate research in India. Indian J Plast Surg 2009;42 Suppl 1:S9-18.

5. Jalili D, Fathi M, Jalili C. Frequency of cleft lip and palate among live births in Akbar Abadi hospital. Acta Med Iran 2012;50:704-6.

6. Scherer NJ, D’Antonio LL, McGahey H. Early intervention for speech impairment in children with cleft palate. Cleft Palate Craniofac J 2008;45:18-31.

7. Sell DA, Grunwell P. Speech results following late palatal surgery in previously unoperated Sri Lankan adolescents with cleft palate. Cleft Palate J 1990;27:162-8.

8. Derakhshandeh F, Rezaei F, Gholmanipur M. Comparative study of the age of receiving therapeutic services in clients referred to Isfahan Cleft Care Team. J Res Rehabil Sci 2011;7:250-8.

9. Kummer AW. Cleft Palate and Craniofacial Anomalies: Effects on Speech and Resonance. 2nd ed. Clifton Park: Singular Thomson Learning; 2008.

10. Shprintzen R, Bardach J. Cleft Palate Speech Management: A Multidisciplinary Approach. St. Louis: Mosby; 1995.

11. Hufnagle K. Therapy techniques for cleft palate speech and related disorders. Cleft Palate Craniofac J 2004;41:340.

12. Peterson-Falzone SJ, Hardin-Jones MA, Karmell MP. Cleft Palate Speech. 4th ed. St. Louis: Mosby/Elsevier; 2010.

13. Vlastos IM, Koudoumnakis E, Houlakis M, Nasika M, Griva M, Styliognianni E. Cleft lip and palate treatment of 530 children over a decade in a single centre. Int J Pediatr Otorhinolaryngol 2009;73:993-7.

14. Austin AA, Druschel CM, Tyler MC, Romitti PA, West II, Damiano PC, et al. Interdisciplinary craniofacial teams compared with individual providers: Is orofacial cleft care more comprehensive and do parents perceive better outcomes? Cleft Palate Craniofac J 2010;47:1-8.

15. Hardin-Jones M, Chapman KL. The impact of early intervention on speech and lexical development for toddlers with cleft palate: A retrospective look at outcome. Lang Speech Hear Serv Sch 2008;39:89-96.

16. Howard S, Lohmander A. Cleft Palate Speech: Assessment and Intervention. Chichester: Wiley-Blackwell; 2011.

17. Tsai TP, Huang CS, Huang CC, See LC. Distribution patterns of primary and permanent dentition in children with unilateral complete cleft lip and palate. Cleft Palate Craniofac J 1998;35:154-60.

18. Brattström V, Mølsted K, Prahl-Andersen B, Semb G, Shaw WC. The Eurocleft study: Intercenter study of treatment outcome in patients with complete cleft lip and palate. Part 2: Craniofacial form and nasolabial appearance. Cleft Palate Craniofac J 2005;42:69-77.

19. Sindet-Pedersen S, Enemark H. Comparative study of secondary and late secondary bone-grafting in patients with residual cleft defects. Short-term evaluation. Int J Oral Surg 1985;14:389-98.