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Original Article

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Evaluating the Frequency of Ear Problems in Patients with Cleft Lip and Palate

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Abstract

Background: Orofacial cleft is one of the most common congenital malformations of craniofacial region. Otitis media with effusion causing conductive hearing loss is a considerable challenge for many children with cleft lip and palate. The aim of this study was to evaluate the prevalence of hearing disorders and associated malformations in these patients.

Methods: The research population consisted of patients with cleft palate, between years 2012 and 2014, who were referred to Children's Medical Center and Vali-e-Asr hospital in Tehran, Iran. Otoscopic examination, tympanometry, pure tone audiometry and echocardiography were performed for each patient.

Results: Among patients with cleft palate, 73% suffered from hearing disorders. There was no relationship between prevalence of hearing loss and sex, presence of other congenital anomalies and degree of cleft, but middle ear diseases were significantly higher in children younger than 2 years. Among patients with cleft lip or palate, 10% suffered from cleft lip, 63% suffered from cleft palate and 27% suffered from cleft lip and palate. There was at least one congenital anomaly in 53% of patients.

Conclusion: This study demonstrates high prevalence of otitis media with effusion and conductive hearing loss in patients with cleft. However, audiologic problems are alleviated when patients become older.

Keywords: Cleft lip, Cleft palate, Hearing disorders, Hearing loss, Otitis media with effusion

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Introduction

Orofacial cleft is one of the most common congenital malformations of craniofacial region (1). The incidence of Cleft Lip and Palate (CLP) varies between 1/500 and 1/2000 live births (2). A child born with cleft lip and palate is more susceptible to various problems such as feeding difficulties, conductive hearing loss, speech problems, dental anomalies and even life threatening anomalies (3). The high prevalence of hearing loss in patients with CLP has been studied for many years. The relationship between middle ear disorders and cleft palate was explained by Alt for the first time (4). The high incidence of Otitis Media with Effusion (OME) in middle ear is mainly due to dysfunction of tensor veli palatini muscles and levator veli palatini muscles and immature development of the Eustachian tube which leads to hearing loss in these patients (5). Several studies have estimated the prevalence of middle ear pathologies to be 50 to 93% in these patients (6). Recurrent otitis media with effusion in the critical growth age can have a profound effect on the learning ability, academic performance and psycho-social development in these children (7). Poor Eustachian tube function can lead to OME even after palatoplasty (8). Thus, many patients need to undergo ventilation tube insertion surgery and repair of cleft palate simultaneously (6). Early diagnosis and proper treatment of middle ear diseases in children with cleft lip and palate can reduce the incidence of hearing impairments and improve the performance of these patients in future.

This study was conducted in order to find out the prevalence of audiological problems in patients with CLP. Effect of age, sex, accompanying anomalies and degree of cleft on the hearing status of children with facial cleft was also assessed.

Materials and Methods

A total of 100 patients with cleft lip and palate were evaluated for auditory status between years 2012 and 2014 in the ENT department of Vali-e-Asr hospital and Children's Medical Center in Tehran, Iran. In this study, all patients with cleft lip, cleft palate or cleft lip and palate younger than 18 years old were included.

All patients were evaluated with the purpose of classifying the degree of lip and palate defects. Classification of degree of cleft palate was as follows: SMCP, submucous cleft palate; SCP, soft cleft palate; USCP, unilateral secondary cleft palate (Posterior to incisive foramen); BSCP, bilateral secondary cleft palate; UPCP, unilateral primary cleft palate (Anterior to incisive foramen); BPCP, bilateral primary cleft palate and revision group including patients that underwent repair of cleft palate prior to entering the study. Also, comprehensive history was taken and complete physical examination was performed in order to evaluate the presence of other congenital anomalies. Sex, age, routine otoscopic examination, tympanometry, pure tone audiometry, conventional behavioral audiometry and echocardiography were recorded. Pure tone audiometry findings for air and bone conduction were obtained for each ear at 250, 500, 1000, 2000 and 4000 *Hz* in a soundproof room. Normal hearing threshold was defined at 20 *dB* or lower. Middle ear pressure and compliance were recorded by using tympanometry. Pressure was considered normal between -100 and+100 *daPa* and was named Type A tympanogram whereas Types B and C were classified as abnormal.

The protocol of this study was approved by the Institutional Review Board of Tehran University of Medical Sciences. Detailed information about the study was given to the participants and a written informed consent was obtained from each one. All aspects of the study were conducted according to the Declaration of Helsinki.

Data were analyzed using IBM SPSS Statistics V21 (IBM, NewYork,USA). Independent T-test and Pearson's chi-squared test were used for analysis. Values were evaluated by using descriptive statistical analysis (Mean±standard deviation), and results were significant at $p<0.05$.

Results

Patients entered into study were in the age range of 1-131 months, with average of 22.25±2.47 months at first examination. There were 55 girls and 45 boys. The age difference between the group of girls and the group of boys was not significant (p=0.593). Table 1 describes the cleft classifications in patients of this study. In total, 10% of cases were rated as cleft lip, 63% as cleft palate and 27% as cleft lip and palate. Thus cleft lip was more frequent in boys whereas cleft palate was more frequent in girls (p=0.035).

Table 1. Cleft classifications

		Total
Cleft palate staging	Submucous cleft palate	6(6.66%)
	Soft palate	12(13.33%)
	Unilateral secondary	5(5.55%)
	Bilateral secondary	34(37.77%)
	Unilateral primary	3(3.33%)
	Bilateral primary	24(26.66%)
	Revision	6(6.66%)

Audiometry and tympanometry were performed for total of 90 patients with cleft palate and cleft lip and palate. Results demonstrated 24 normal cases (11 boys and 13 girls) and 66 abnormal cases (26 boys and 40 girls). In other words, the prevalence of abnormal audiometric results was 70% in males and 75% in females but the difference was not significant (p=0.583). Table 2 demonstrates the results of audiometry test.

In addition to audiometry, routine otoscopic examinations were performed for the 66 patients who had abnormal results in audiologic evaluations. Statistical analysis showed no difference between genders in otoscopic examinations. Among 66 patients with abnormal audiologic examination, 3 patients appeared to be normal in microscopic examination (OR) and 63 patients required myringotomy. Moreover, 52 patients needed ventilation tube insertion and this surgery was not necessary for 11 patients due to absence of glue ear. Each one of unilateral aural atresia and bilateral aural atresia were present in 1 patient, both patients suffered from bilateral secondary cleft palate. Results of otoscopic examinations are summarized in tables 3 and 4.

There was hearing loss in 75% (47 of 63) of patients

Table 2. Results of audiometry and tympanometry in patients with cleft palate and cleft lip and palate (p=0.583)

		Male	Female	Total
Tympanometry	Unilateral B/C type	4	8	12(14.81%)
	Bilateral B/C type	22	32	54(60%)
	Total	26	40	66(73.33%)
Audiometry	Unilateral CHL*	4	8	12(14.81%)
	Bilateral CHL	21	31	52(57.77%)
	Mixed CHL	1	1	2(22.22%)
	Total	26	40	66(73.33%)

* Conductive hearing loss

Table 3. Results of otoscopic exam by sex

Otoscopic exam		Normal	Unilateral serous otitis media	Bilateral serous otitis media
Sex	Male	11(12.22%)	5(5.55%)	21(23.33%)
	Female	13(14.44%)	6(6.66%)	34(37.77%)

Table 4. Results of otoscopic exam at different stages of cleft palate

Otoscopic exam results		Normal	Unilateral serous otitis media	Bilateral serous otitis media
Cleft palate staging	Submocusal cleft palate	3(3.33%)	0	3(3.33%)
	Soft	6(6.66%)	2(2.22%)	4(4.44%)
	Unilateral secondary	0	0	5(5.55%)
	Bilateral secondary	10(11.1%)	2(2.22%)	20(22.2%)
	Unilateral primary	0	0	3(3.33%)
	Bilateral primary	5(5.55%)	3(3.33%)	16(17.7%)
	Revision	3(3.33%)	2(2.22%)	1(1.11%)

with cleft palate and in 70% (19 of 27) of patients with both cleft lip and palate. Children with primary type of cleft palate were at higher risk of hearing impairments compared to the secondary type. Moreover, the incidence of middle ear disease was 81% (22 of 27 patients) in children with primary type and 73% (27 of 37) in children with secondary type but this difference was not statistically significant (p=0/497). Furthermore, children younger than 2 years of age were significantly at higher risk of developing middle ear disease than those older than 2 years of age (p=0.022).

Discussion

Orofacial clefts are one of the most prevalent congenital malformations in the craniofacial area (1). Various genetic and environmental factors including alcohol consumption, smoking and antiepileptic medication play a role in etiology and pathogenesis of this disease (2). Hearing difficulties as a result of otitis media with effusion can have a detrimental effect on cognitive and educational performance of these children in the future (7).

A total of 100 patients with cleft lip or palate who were referred to the Children's Medical Center and Vali-e-Asr hospital in Tehran between years 2014 and 2015 were included. Among these patients, 10% were rated as cleft lip, 63% as cleft palate and 27% as cleft lip and palate, which is almost consistent with findings of Shafi *et al* who reported 19% for cleft lip, 45% for cleft palate and 36% for cleft lip and palate among 123 patients in their study (9). Yazdee *et al* conducted a study on 177 similar patients in Iran and reported 25.4% for cleft lip, 22.6% for cleft palate and 52% for cleft lip and palate (10).

In our study population, the rate of cleft lip was higher in males and cleft palate in females. The higher prevalence of cleft palate in females could be due to delay in the formation of palate. These results are in line with the findings of Ingalls *et al*, Czeizel *et al*, Shafi *et al*, and Sayetta *et al* (11-14). In the study of Yazdee *et al,* cleft lip and palate was more prevalent in males but cleft lip was more frequent in females and cleft palate was more frequent in males (10).

The prevalence of hearing disorders in patients with cleft lip and palate is higher than the normal population. This high prevalence is attributable to dysfunction of tensor veli palatini muscles and levator veli palatini muscles and immature development of the Eustachian tube (5). The high incidence of auditory disorders in children during their growth ages can lead to impairments in educational, cognitive and linguistic abilities and interfere with their psycho-social development (7). Our data indicated that the prevalence of auditory disorders is high in patients with cleft lip and palate which is in accordance with other studies. In a study carried out by Viswanathan *et al*, 90-96% of patients suffered from otitis media with effusion or conductive hearing loss which was mostly bilateral (15). Luthra *et al* reported unilateral otitis media with effusion in 25 patients among the total of 55 patients and bilateral otitis media with effusion in 16 patients. In a study by Flyn *et al* among 81 patients, 61 suffered from otitis media with effusion (16).

Generally, 75% of patients with cleft palate and 70% of patients with cleft lip and palate demonstrated auditory disorders. Similar to Gani *et al*'s findings, our study indicates that there is no association between the prevalence of auditory disorders and type of the cleft (24). However, other studies show contradicting results (25). In a study by Schwartz *et al*, patients with milder clefts suffered from lower risk of developing otitis media which emphasizes the role of cleft palate severity in ensuing complications. Luthra *et al* indicated a significant association between hearing loss and tympanometric findings with cleft type (16). Other studies found no association between cleft type and auditory outcomes such as hearing loss, otitis media and number of surgeries for ventilation tube insertion (19-23).

In our study, 70% of boys and 75% of girls had abnormal otoscopic examinations. The difference was not significant and demonstrates a lack of association between sex and auditory disorders in CLP patients. Sixty-eight patients were younger than 2 years and 22 patents were older; among them, 79% and 54% suffered from hearing loss, respectively. The difference between these two age groups was statistically significant. A study conducted by Chu and McPherson and another one by Ma *et al* showed no association between age, sex and cleft type with the findings of auditory examination (1,24). In a study by Gould *et al*, cleft type and sex were not associated with auditory status but there was a

significant association between ethnicity and age with the prevalence of auditory disorders which decrease in older patients (25).

Conclusion

Our data indicates that auditory disorders are not associated with sex or the presence of an accompanying anomaly but the prevalence of these disorders extremely decreases when the patients grow older.

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