



Original Article

The Prevalence of Cleft Lip and Palate Patients: A Single-Center Experience for 17 Years

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ABSTRACT

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Objective: The aim of the present study was to report the prevalence of cleft lip and palate (CLP), isolated cleft palate (CP), isolated cleft lip (CL), and median cleft (MC) applied in Marmara University School of Dentistry, Department of Orthodontics.

Methods: There were 1058 patients who applied to the center between 2000 and 2017. A total of 1026 patients were included in the study. Files, models, and photographs were evaluated. The patients were divided into six groups: unilateral right or left CLP, bilateral CLP, isolated CP, isolated CL, and MC. The prevalence was identified according to cleft type, side, gender, age, and application year.

Results: The most common type was unilateral CLP (44.3%), which was observed more on the left side (28.9%), followed by CP (28.7%). MC had a minimum ratio (0.3%) between cleft types. Males were more prone to have unilateral CLP (right side, 64.6%) and bilateral CLP (64.1%), whereas females were more prone to have CP (59.9%). The greatest number of applications was recorded in 2004, and the patients mostly applied in the neonatal period (64.9%). The ratios of complete cleft cases in all types of clefts were statistically significantly higher.

Conclusion: Unilateral CLP was the most common cleft type and seen more on the left side. While males were affected more by CLP, isolated CP was seen more in females than in males.

Keywords: Cleft lip and palate, prevalence, single center

INTRODUCTION

Cleft lip and palate (CLP) is one of the most prevalent malformations occurring in the head and neck region. The etiology of this malformation is multifactorial, and the incidence of clefts may be affected by ethnic, racial, geographic, and socioeconomic factors (1). Current knowledge indicates that orofacial clefts occur in approximately 1 in 700 live births, and that 3200 new cases per year are expected with the population growth worldwide (2, 3).

For the epidemiology of CLP, there are well-designed overviews, which include regional data from the European Registration of Congenital Anomalies and Twins, International Clearinghouse for Birth Defects Monitoring Systems, and National Birth Defects Study. However, Turkey is not included in those groups and does not have any national data showing the prevalence of CLP cases. A few studies were conducted in different regions of Turkey (Ankara (2, 4), Denizli (5), and Konya (1)) reporting the ratio and relationship of CLP with other malformations. Therefore, current data are insufficient for prevalence of CLP in Turkey.

According to national records, almost 30% of the whole population in Turkey lives in Istanbul. Two state universities and two private universities apply preoperative treatment to newborns with CLP in Istanbul regularly. A large number of the patients come from neighboring/other cities to Istanbul and to our clinic to receive preoperative treatment.

The aim of the present study was to report the prevalence of CLP (unilateral or bilateral), isolated cleft palate (CP), isolated cleft lip (CL), and median cleft (MC) patients who applied in Marmara University School of Dentistry, Department of Orthodontics for treatment.

METHODS

The present study was conducted by using the records in the cleft archive of Marmara University School of Dentistry, Department of Orthodontics. According to registration records, a total of 1058 patients applied to the clinic between 2000 and 2017. The routine protocol of our clinic for clefts consisted of filling out a special CLP form (including information about birth date, weight, height, cleft type, birth type, name of the gynecologist, parents' ages, profession, birthplace, smoker or nonsmoker, any drug usage, radiation exposure, number of children, and presence of cleft in the family), extraoral and intraoral photographs, and impressions. Informed consent was obtained from all patients or their parents for use of their records.

Files, stone/digital models, and photographs of 1026 patients were evaluated. The patients were divided into six groups: unilateral right or left CLP, bilateral CLP, isolated CP, isolated CL, and MC. The prevalence was classified according to cleft type, cleft side, gender, and subgrouped according to years and ages.

Exclusion criteria were patients who refused to fill out the CLP forms, clefts combined with syndromes, and clefts with undefined and insufficient information.

The Statistical Package for Social Sciences version 22.0 program (IBM Corp.; Armonk, NY, USA) was used for statistical analysis. Chi-square analysis was used for comparisons. A $p < 0.05$ was considered as significant. When the total number of cases in some categories was insufficient, these categories were removed from the study and then compared for cross tabulation by using Chi-square analysis.

Table 1. Distribution of gender

	Frequency (n)	%
Boys	556	54.2
Girls	470	45.8
Total	1026	100

Table 2. Type of cleft

	Frequency (n)	%	Valid %	Cumulative %
Bilateral CLP	262	25.5	25.5	25.5
CP	294	28.7	28.7	54.2
CL	12	1.2	1.2	55.4
MC	3	0.3	.3	55.7
Unilateral CLP-right	158	15.4	15.4	71.1
Unilateral CLP-left	297	28.9	28.9	100.0
Total	1026	100.0	100.0	

RESULTS

The number of males (n=556; 54.2%) who applied to the university with chief complaints of cleft was higher than that of females (n=470; 45.8%) (Table 1).

The most common type of cleft was unilateral CLP with a ratio of 44.3% (n=455 patients), followed by isolated CP with 28.7% (n=294 patients) (Table 2). Unilateral CLP was seen more on the left side with a ratio of 28.9% (n=297 patients) than on the right side regardless of gender difference. Bilateral CLP had a ratio of 25.5% (n=262 patients). Median (n=3; 0.3%) and lip (n=12; 1.2%) clefts presented the minimum ratios.

Regarding the application period for treatment need, it was seen that 64.9% of the patients (n=666) applied in the neonatal period (0-1 year) (Table 3). The second highest ratio recorded was 11.31% (6-10 years), and the lowest ratio was recorded after 30 years old.

The greatest number of applications was recorded in 2003 (n=110 patients) and 2004 (n=118), whereas the minimum number (n=20) was in 2015 (Table 4). The distribution of the patients according to years, gender, and cleft types is shown in Table 4.

Since lip and MC types showed inadequate sample numbers, statistical comparisons were performed by chi-square tests after these categories were excluded.

Comparisons regarding cleft type and gender showed that bilateral CLP was seen to be statistically significantly higher in boys (64.1%); however, isolated CP was seen to be statistically significantly higher in girls (59.9%). Unilateral right-sided CLP was seen to be significantly higher in boys (64.6%), whereas there was no statistically significant difference between the genders with regard to unilateral left-sided CLP (Table 5).

The ratios of complete cases in unilateral and bilateral cleft types were statistically significantly higher than those of incomplete cleft types (Table 6).

DISCUSSION

The racial prevalence is highest in Whites, followed by Hispanics, Asians, and Africans, respectively (6, 7). The national US average

Table 3. Distribution of ages

Age (years)	n	%
<1	666	64.9
1-5	99	9.65
6-10	116	11.31
11-15	75	7.31
16-20	46	4.48
21-25	15	1.47
26-30	5	0.49
>30	4	0.39

Table 4. Distribution of the patients according to application years

Years	Total	Male							Female						
		Total	BL	Palate	Lip	Median	UL—right	UL—left	Total	BL	Palate	Lip	Median	UL—right	UL—left
2000	47	25	10	3	1	0	5	6	22	6	5	0	0	6	5
2001	27	16	7	2	1	0	1	5	11	2	4	0	0	0	5
2002	66	45	13	17	0	0	7	8	21	4	6	1	0	4	6
2003	110	56	20	11	2	0	7	16	54	16	18	0	1	4	15
2004	118	65	16	13	0	0	9	27	53	9	16	0	0	7	21
2005	81	45	17	6	0	0	7	15	36	9	11	1	0	3	12
2006	71	39	11	4	1	0	11	12	32	4	11	1	0	5	11
2007	71	42	14	9	1	0	9	9	29	5	10	0	0	7	7
2008	71	33	11	5	0	0	10	7	38	7	13	0	0	1	17
2009	52	26	4	11	0	0	7	4	26	7	14	0	1	1	3
2010	55	30	7	6	0	0	6	11	25	3	12	1	0	3	6
2011	41	21	7	4	0	0	5	5	20	4	6	0	0	4	6
2012	49	22	6	4	1	0	5	6	27	3	15	0	0	3	6
2013	40	25	10	2	0	1	5	7	15	5	6	0	0	1	3
2014	42	25	7	4	0	0	4	10	17	3	6	0	0	3	5
2015	20	9	3	1	1	0	2	2	11	2	6	0	0	1	2
2016	31	16	3	10	0	0	1	2	15	2	7	0	0	1	5
2017	34	16	2	6	0	0	1	7	18	3	10	0	0	2	3

Table 5. Comparisons Regarding Cleft Type and Gender

			Gender		
			Male	Female	Total
CLEFT TYPE	Bilateral CLP	Count	168 _a	94 _b	262
		% Within cleft type	64.1%	35.9%	100.0%
		% Within gender	30.7%	20.3%	25.9%
	CP	Count	118 _a	176 _b	294
		% Within cleft type	40.1%	59.9%	100.0%
		% Within gender	21.6%	37.9%	29.1%
	Unilateral CLP-right	Count	102 _a	56 _b	158
		% Within cleft type	64.6%	35.4%	100.0%
		% Within gender	18.6%	12.1%	15.6%
	Unilateral CLP-left	Count	159 _a	138 _a	297
		% Within cleft type	53.5%	46.5%	100.0%
		% Within gender	29.1%	29.7%	29.4%

There is a statistically significant difference between the genders indicated by small letters in the same line (p <0.05); Chi-square tests

Table 6. Comparison of complete/incomplete cases in unilateral and bilateral cleft types

			Total	Complete	Incomplete
Cleft type	Bilateral	Count	231 _a	29 _b	260
		% within cleft type	88.8%	11.2%	100.0%
	Unilateral-right	Count	131 _a	27 _b	158
		% within cleft type	82.9%	17.1%	100.0%
	Unilateral-left	Count	230 _a	67 _b	297
		% within cleft type	77.4%	22.6%	100.0%

There is a statistically significant difference between the complete/incomplete cases indicated by small letters in the same line (p <0.05); Chi-square tests

rate was 7.75% with the highest value in Maryland (21.46%), and the lowest was found in West Virginia (2.59%) (8). American In-

dians had the highest ratio (9), and African-Americans had the lowest ratio from 0.21 to 0.41 per 1000 live births (10). Whites

in Western Europe and the United States had an incidence rate ranging from 0.77 to 1.40 per 1000 live births (9).

Asian countries demonstrate close ratios. The incidence rates were from 1.14 to 2.13 per 1000 live births in Japanese and 1.81 per 1000 or 1 in 554 live births in South Koreans (11, 12). Murray and Martelli-Junior (13, 14) have reported the incidence rates to be 1.94 and 1.46 per 1000 live births in the Philippines and Brazil, respectively. In Caucasians, the incidence for CL with or without palate was between 0.6 and 1.7 per 1000 live births (2, 3).

In the literature, it was reported that an average of 66.9% of the affected children had CLP. In our study, the most common cleft type was also CLP (69.8%). However, the average ratio for CLP may increase or decrease according to the regions (Mexico–South America and USA have a higher proportion, 66.9% and Eastern Europe, the British Isles, and South-Mediterranean Europe have lower proportion) (6). CLP was seen in higher ratios in Latin American and Asian (China and Japan) populations (15), thus coinciding with the results in our department. Furthermore, isolated CP was reported in higher ratios in Canada and Northern Europe, which was in contrast to our ratio (6).

Literature reviews reported that CLP tends to be unilateral and occurs more frequently on the left side (16). The International Perinatal Database of Typical Oral Clefts study results showed that 30.2% of the CLP group had bilateral cleft and 69.8% had unilateral cleft. The defect ratios were 41.1% on the right side and 58.9% on the left side (6). Although the laterality was the same as our study, the percentage was slightly higher (65.27% on the left side in our study).

CL with or without CP was seen more often in males; however, CP was seen more frequently in females (17). Van den Akker (18) and Stoll (19) found that boys appear to be affected more in bilateral cases, thereby coinciding with our results. On the other hand, Meskin (20) and Henriksson (21) reported that girls had bilateral CL more than boys.

When we overlooked the studies regarding the incidence rate of CL with or without palate in Turkey, it was reported to be 0.95 per 1000 births, and the ratio of isolated CP was reported to be 0.77 per 1000 births (4). According to the study in Hacettepe University, Ankara, Turkey, 64.4% of the patients had CL with or without CP, and 35.6% had isolated CP (4). Altunhan et al. (1) published a study about the incidence of congenital anomalies associated with CP and CLP in Konya region and found that 71% of the patients have CLP, and that in 80% of the patients, only one side is affected. Another study including Denizli region reported 65.5% CP, 21.6% CL, and 12.9% CLP ratios between 2004 and 2010 (5).

Gender differences also exist for CLP. Boys are affected more often and have more severe clefts than girls (22, 23). However, girls are affected more often with isolated CP than boys. In Ankara, Turkey, Borçbakan (2) reported a study with 1000 patients between 1955 and 1965 and found that males appear to be affected more; however, they explained that more males applied to their clinic.

In our study, the frequency distributions were 69.8% for CLP and 28.7% for isolated CP. In Hacettepe University's study, 64.4% of the patients had CL with or without CP, and 35.6% had isolated CP, thus coinciding with our results although the ratios showed minimal difference (4). Our study results also coincided with the study by Altunhan et al. (1) in Konya region. Although both studies showed that unilateral CLP was more often, the ratio for unilateral CLP was reported to be 17.3%, which was significantly lower than the ratio in our study. However, males were found to be affected more in our study, which was a controversial result in Konya region's study (1).

In 2013, Tomatir et al. (5) reported that isolated CP is seen more frequently (65.5%), which did not coincide with our results (28.7%). However, the results were almost similar regarding genders. Both studies showed that males are affected by clefts more often. Previous studies showed that isolated CP was seen more frequently in girls. While our results coincided with the literature, Borçbakan et al. (2) in 1969 reported that all types of clefts are seen more often in males; however, they explained that their results have more male than female patients. They also found that unilateral CLP has a higher ratio, thereby coinciding with our results.

One of the disadvantages of these studies might be the lack of standardization in classification. While Borçbakan (2) and Tomatir (5) classified their patients as CL, CP, and CLP, Altunhan (1) and Tunçbilek (4) classified their patients as CP and CLP. Furthermore, Altunhan (1) and Borçbakan (2) divided the patients into two groups as unilateral and bilateral clefts. However, only Borçbakan (2) subgrouped the patients according to laterality as right and left sides. Therefore, the patients were divided into as many subgroups as possible in our study.

An increase in the number of patients who applied was observed between 2000 and 2005, which might be related with the application of nasoalveolar molding (NAM) that became widespread, and the success of this treatment protocol was approved. Furthermore, surgeons started to refer more patients to the orthodontists because of the positive effects of NAM therapy. The reason for the steadily descending number of applicants after 2005 might be explained by other faculties that started to treat CLP patients in many other cities every year, suggesting that many patients applied to the faculties closer to them. In addition, in 2015, there is a critical decrease in the number of applicants because our faculty moved from the original campus to the new one in another county, and thus we were unfortunately unable to accept new patients during this period. They were all referred to other hospitals. After the settlement was finished, the number of CLP applicants started to increase; however, since many other faculties treat CLP patients nowadays, it is thought that the number will never increase as in the previous years. On the other hand, the treatment of patients with CLP requires experience. Therefore, such patients should be treated by specialized and experienced individuals in CLP centers.

On the other hand, when we examined the application period for treatment need, there was a negative correlation between

ages and application numbers, suggesting that adult applicants are not motivated to seek for orthodontic treatments. This might be also explained by the reason that the patients usually continue their orthodontic treatments in the same university, where they applied for presurgical infant orthopedics during the first months of their lives. Therefore, 64.9% of the patients who applied to our clinic for treatment were in the neonatal period (0–1 year). Second, there was an increase between the ages of 6 and 10 years (11.31%), which is the period of mixed dentition that malocclusions become more pronounced and primer orthodontic treatments are started, cross bites were corrected, neighboring teeth to the cleft area were leveled, orthopedic treatment, such as facemask, was applied, and bone grafting was performed.

Only few studies grouped the patients as complete or incomplete. Sivertsen et al. (24) reported that the ratio of complete cases (81%) is significantly higher than that of incomplete cases. In their study, 88% of the patients had bilateral complete CLP, which is very similar to the ratio found in our study (88.8%). In addition, they reported that 77% of the patients have unilateral complete CLP, whereas they found no significant difference in laterality. Carroll et al. (25) only included unilateral CLP cases and found that 88.8% of the patients have unilateral complete CLP and no significant difference in laterality. In our study, although the patients were divided into two as right and left unilateral CLPs, the ratios were similar (82.9% and 77.4%, respectively). Martelli-Junior et al. (14) reported complete unilateral CLP as the most prevalent cleft type with a ratio of 26.19% between all cleft types. When complete cases were evaluated in-between, unilateral CLP was found to be 66%, and bilateral CLP was found to be 34%. In another study from Brazil, Freitas et al. (26) reported that the most common cleft type is complete CLP (37.1%), similar to our study. Conversely, only Shapira et al. (27) found that incomplete CLP is the most frequent type of cleft (71%).

CONCLUSION

Unilateral CLP was the most common cleft type applied for treatment in Marmara University, Faculty of Dentistry, Department of Orthodontics, and most of the patients applied in the neonatal period (0-1 year). Furthermore, unilateral cleft was seen more frequently on the left side. Males were affected more by CLP, and isolated CP was seen more in females than in males. The ratios of complete cases were statistically significantly higher than those of incomplete cleft types. While the number of applicants increased between 2000 and 2004, over the years, the number started to decrease because of new faculties.

Although Istanbul might be considered as a preferable city for reflecting a general data about the prevalence of CLP in Turkey because of the reasons mentioned above, it still does not reflect a clear data for whole Turkey. In fact, it would be better to conduct that kind of study with collecting data from those three universities. Furthermore, comprehensive national studies are needed to assess the real national data.

Ethics Committee Approval: Ethics committee approval was received for this study from the Ethics Committee of Marmara University.

Informed Consent: Written informed consent was obtained from the patients and the parents of patients who participated in this study.

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