

The distribution and classification of clefts in patients attending a cleft lip and palate clinic in Riyadh, Saudi Arabia

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ABSTRACT

الأهداف: تقييم ومناقشة مسببات شق الشفة وقبة الحنك CLP، وتوزيعها وتصنيفها عند المرضى المراجعين لواحدة من أنشط عيادات شق الشفة وقبة الحنك CLP.

الطريقة: جمعت جميع سجلات 171 مريضاً المرجعين لعيادة شق الشفة وقبة الحنك CLP بكلية طب الأسنان، جامعة الملك سعود في مدينة الرياض – المملكة العربية السعودية، وذلك في الفترة من نوفمبر وحتى ديسمبر 2006م. استخدم اختبار Z النسبي للتحليل الإحصائي للمتغيرات المسببات، والعمر، والجنس، ونوع الشق وموقعه ($p < 0.05$).

النتائج: تراوحت أعمار المرضى ما بين نصف سنة إلى 30 سنة، بمتوسط 11.1 سنة، (انحراف معياري 5.87). كان معدل الانتشار بين المرضى المراجعين الذكور (63.7%) أكثر من المرضى الإناث (36.6%) بدلالة إحصائية ($p = 0.000$). أظهرت النتائج أن العامل البيئي المجهول هو من أكثر العوامل المسببة للمرض (79.5%) بدلالة إحصائية ($p = 0.000$). شق الشفة والحنك CLP كان من أكثر النماذج انتشاراً (78.3%) بدلالة إحصائية ($p = 0.000$), ضمن مختلف أنواع الشقوق وضمن الشق نفسه كان شق الشفة وقبة الحنك أعلى بشكل واضح عند الذكور (49.1%) (62.7%) مقارنة بالإناث (29.1%) (37.3%) وبدلالة إحصائية ($p = 0.008$) ($p = 0.038$) على التوالي. ولم يلاحظ أي اختلاف إحصائي جوهري بين الجانبين الأيمن والأيسر.

خاتمة: متوسط العمر ومداه عند مرضى شق الشفة وقبة الحنك CLP عند الزيارة الأولى كان مرتفعاً. المرضى الذكور أكثر من الإناث. لم يتم تحديد العامل البيئي المسبب. يعتبر نوع شق الشفة والحنك CLP من أكثر نماذج شقوق الفم شيوعاً، نسبته أعلى في الذكور عن الإناث.

Objective: To evaluate and discuss the etiology, distribution, and classification of cleft lip and palate (CLP) patients in one of the most active CLP clinics in Riyadh, Saudi Arabia.

Methods: The records of all 171 CLP patients attending the CLP clinic at the College of Dentistry, King Saud University in Riyadh, Saudi Arabia, were collected in November and December 2006. Variables such as etiology, age, gender, type of cleft, and location of cleft were statistically analyzed. Proportional Z test ($p < 0.05$) was used.

Results: The patients' age range was 0.5-30 years with an average of 11.1 (standard deviation 5.87) years. Attending male patients (63.7%) were more than females (36.6%) ($p = 0.000$). The unknown environmental factor was the most reported etiological factor (79.5%) ($p = 0.000$). The cleft lip and palate were the most prevalent type (78.3%) ($p = 0.000$). In the different types of cleft, as well as within the same cleft type, the cleft lip and palate type were significantly higher in males (49.1%) (62.7%), than females (29.1%) (37.3%) with ($p = 0.038$) ($p = 0.008$). There were no statistical differences between the right and left cleft sides.

Conclusions: The average age and range of CLP patients at first visit were high. Attending male patients was more than females. The environmental etiological factor was not identifiable. Males were dominant in the cleft lip and palate type. The cleft lip and palate type were the highest among oral clefts.

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Orofacial clefting including cleft lip with or without cleft palate (CLP) is considered one of the most common craniofacial anomalies. There appear to be an increase in the frequency of congenital malformation, with orofacial clefting representing 15% of all such anomalies.¹⁻⁴ Epidemiologic data on clefting may be of limited value if all types of orofacial clefts were combined without distinction between the different types of clefts, location, etiology, classification, severity, and so forth. This information is also important for the successful management of cleft lip and/or cleft palate patients. Such management is best performed by a team of clinicians who need to have a thorough knowledge of oral clefts.¹ Several papers have been published in regard to orofacial clefts in Saudi Arabia and the Arab population.⁵⁻¹⁵ However, few articles reported and discussed incidence, type, location, severity, gender differences in cleft patients attending treatment in Saudi Arabia.^{7,8,10} Therefore, the present study was undertaken to investigate the possible etiology, distribution and classification of clefts in patients attending the cleft lip

and palate clinic at the College of Dentistry, King Saud University, Riyadh, Saudi Arabia.

Methods. After the approval of the College of Dentistry Research Center ethical committee to conduct this retrospective study, the records of all 171 CLP patients attending the CLP clinic (Orthodontic clinic) at the College of Dentistry, King Saud University, Riyadh, Saudi Arabia, were collected between November and December of 2006 and evaluated. Variables including etiology, age, gender, type of cleft, and location of cleft, were all investigated using descriptive statistics, and the analytical proportional Z test ($p < 0.05$) provided by the SPSS 12.0 software Window's Program (SPSS Inc., Chicago, Illinois, U.S.A.).

Results. The patients' age range was 0.5-30 years with an average of 11.1 (standard deviation 5.87) years. Attending male patients (109, 63.7%) were more than females (62, 36.6%) ($p = 0.000$). The unknown environmental factor (79.5%) was the most reported etiological factor followed by the unknown genetic factor (16.4%) ($p = 0.000$), **Table 1**. The cleft lip and palate was the most prevalent type (78.3%, $p = 0.000$). In the different types of clefts, as well as within the same cleft type, cleft lip and palate was significantly higher in males (49.1%) (62.7%), than females (29.1%) (37.3%) with ($p = 0.038$) and ($p = 0.008$). There was no statistical difference between the right and left cleft sides in all cleft types, **Table 2**.

Discussion. This study may help in providing more clinical information about CLP patients in Saudi

Table 1 - Etiological factors.

| Sub-heading | No. | (%) |
|----------------------|------------|----------------|
| <i>Genetics</i> | | |
| Father side | 1 | (0.6) |
| Mother side | 1 | (0.6) |
| Unknown | 28 | (16.4) |
| <i>Environmental</i> | | |
| Unknown | 136 | (79.5)* |
| Drugs | 4 | (2.3) |
| Radiation | 1 | (0.6) |
| Total | 171 | (100.0) |

* $p = 0.000$

Table 2 - Distribution of clefts according to side and gender.

| Type of cleft | Right side | Left side | Median | Bilateral | No. of male/ female within the cleft type (%) | No. of male/ female in different cleft types (%) | No. of combined male & female in different cleft type (%) |
|-------------------------|------------|-----------|-----------|-----------|--|--|--|
| <i>Lip</i> | | | | | | | |
| Male | 5 (45.5) | 5 (45.5) | 0 (0.0) | 1 (9.1) | 11 (64.7) | 11 (6.4) | 17 (9.9) |
| Female | 1 (16.7) | 2 (33.3) | 0 (0.0) | 3 (50.0) | 6 (35.3) | 6 (3.5) | |
| <i>Lip and alveolar</i> | | | | | | | |
| Male | 2 (28.6) | 4 (57.1) | 0 (0.0) | 1 (14.3) | 7 (70.0) | 7 (4.1) | 10 (5.9) |
| Female | 0 (0.0) | 3 (100.0) | 0 (0.0) | 0 (0.0) | 3 (30.0) | 3 (1.8) | |
| <i>Lip and palate</i> | | | | | | | |
| Male | 21 (25.0) | 25 (29.8) | 0 (0.0) | 38 (45.2) | 84 (62.7)* | 84 (49.1)† | 134 (78.3)‡ |
| Female | 11 (22.0) | 17 (34.0) | 0 (0.0) | 22 (44.0) | 50 (37.3) | 50 (29.2) | |
| <i>Isolate palate</i> | | | | | | | |
| Male | 0 (0.0) | 0 (0.0) | 7 (100.0) | 0 (0.0) | 7 (70.0) | 7 (4.1) | 10 (5.9) |
| Female | 0 (0.0) | 0 (0.0) | 3 (100.0) | 0 (0.0) | 3 (30.0) | 3 (1.8) | |

* $p = 0.038$, † $p = 0.008$, ‡ $p = 0.000$

Arabia, especially with the very limited published papers.^{7-10,15} The reported age in this study of the 171 cleft patients was the age when the patients were first seen in our cleft lip and palate clinic. Late presentation is rare in the West, however, in developing countries it is not uncommon to find adult unoperated cases. Our age range (0.5-30 years) was extremely wide and even wider than the 6-26 years of the 63 cleft patients reported by AlKofaidi and Barakati in 2002,¹⁰ obtained from this same cleft lip and palate clinic. The age diversity may indicate that there is an increase in the awareness of patients/parents, and/or an increase in the demand for such treatment. This may also be supported by the fact that, patients attending the same clinic almost tripled from 63 patients in 2002 to 171 patients at the end of 2006. The attending male patients (63.7%) were more than females (36.6%) ($p=0.000$). The mode and the mean, which were 8 years and 11.1 (SD 5.87) years are within the usual recommended age for expansion of the collapsed buccal segments as well as for the alveolar bone grafting of the clefts. This may indicate the need to consider time, efficiency, and care in performing buccal expansion and the following alveolar bone grafting by the treating team clinicians, since there is very little spare time between first seeing the patient and the time to perform buccal expansion and alveolar bone grafting for most those patients. Kumar⁷ reported that most his patients 67% were first seen before the age of 2 years. However, 23.5% were between the age of 2-10 years, and 7.8% were after the age of 10 years. Kumar⁷ collected his data from patients attending the Plastic Surgery Department at King Khalid University Hospital, whereas the data of our study were obtained from the orthodontic clinic treating cleft lip and palate patients (CLP clinic), at the College of Dentistry. This logically may explain the higher age reported in our study compared to that in his study. The etiology of cleft lip and palate is considered by researchers as multi-factorial. The expression of a multi-factorial disorder for cleft lip and palate is thought by some researchers to be based on a threshold model, whereby those with more of the disease causing genes and environmental factors as teratogens, are more likely to exceed the threshold and express the disorder.¹⁶ The impact of a teratogen to cause cleft lip or palate depends on the genotype of the mother and the child and the timing and the dose of the drug.¹⁷ Therefore, oral clefts are either genetic in origin or environmental with the consensus that hereditary is the most significant etiology of clefts.¹⁶ This study was not designed to investigate the etiological factors (genetic and/or environmental teratogens) in depth as reported by some,¹⁶⁻²³ nor was

it design to verify the role of consanguinity as reported by others.^{8,24-28} It was simply designed to give an idea of the possible etiological factor, whether genetic or environmental. However, even with this simplified concept, the information that was obtained from the patients and/or parents was not decisive or conclusive. In most cases (79.5%), the "unknown" environmental factor was the possible etiological factor. That is, in those cases the patients and/or parents expressed that both their close and distant family members did not exhibit any form of cleft lip and/or palate and at the same time they could not link the existed cleft to specific drug administration or radiation exposure. This was followed by the "unknown" genetic factor (16.4%), were the patients and/or parents denied that their parents and/or their relatives (grandfathers/mothers, uncles, aunties, and so forth.) have any form of clefts, while the patient did have brothers and/or sisters with clefts, and again with no possible explanation or link of any environmental factors by the patients or their parents. With such contradicting informations, the identification of the etiological factor was not possible. In our study, the most prevalent type was the cleft lip and palate (78.4%) ($p=0.000$), which was in agreement with several studies.^{1,6,7,10,11,13,14,29} However, some of those studies mentioned their-higher-prevalence without conforming it statistically. In the different types of clefts, as well as, within the same cleft type, the cleft lip and palate type was significantly higher in males (49.1%) (62.7%) than females (29.1%) (37.3%) with ($p=0.038$) and ($p=0.008$). This was in contrary to some,¹⁴ and in agreement with others,^{1,7,8,10,13} with the exception of isolated cleft palate. Again in some of these studies, such findings were not statistically confirmed. Even though our findings show the left sides were more affected than the right sides percentage wise. However, such findings were not statistically significant, which was in agreement with some,¹³ and disagreement with others.^{1,7,8,10} This study was not an epidemiological study representing the whole Saudi Arabian population. It was mainly limited to provide general information of the possible etiology, distribution, and classification of CLP patients attending treatment in an active CLP clinic in Saudi Arabia.

In conclusion, the age of cleft lip and palate patients seen in our clinic was relatively large and with very wide range. Attending male patients was more than females. Even though the environmental factor appeared to be highly related to the etiology, however, the environmental factor was not identifiable. Cleft lip and palate was the most common type of cleft. Male patients were more than females in the cleft lip and palate type.

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