

ORIGINAL RESEARCH

A Preliminary Report on Cleft Deformities of the Face and associated Anomalies in Abha, Saudi Arabia

Rafi A Togoo, Syed Mohammad Yaseen, Zakirulla Meer, Ahmad AlMohy, Jaber AlQahtani

ABSTRACT

Cleft deformities of the face are one of the major congenital anomalies seen in our environment. There is a dearth of data from this major Southern city of Saudi Arabia on the incidence of the deformities and associated anomalies. This preliminary report aims to record the pattern of cleft lip/palate and associated anomalies at Aseer Central Hospital in Abha, Saudi Arabia.

Materials and methods: A retrospective study was conducted at Aseer Central Hospital, Abha, to identify all cleft lip and palate patients that reported or were treated between 2005 and 2011. Cleft lip and/or palate records were obtained from patient's files in Department of Maxillofacial Surgery, Pediatrics and Medical records. Gender of patient, type of cleft and any associated congenital anomalies/syndromes were recorded.

Results: Of the patient records studied, 60% were patients male and 40% female. Isolated cleft palate was the most common at 60% followed by combined cleft lip palate at 24% and isolated cleft lip at 16%. Eleven percent of the patient's studies showed associated anomalies with isolated cleft palate.

Conclusion: The study concluded that isolated cleft palate cases are more common than other variants of orofacial clefts. The pattern of cleft observed in the current study was similar to reported studies for Arab populations. This preliminary report lays the foundation for large population and birth registry based studies for prevalence and frequency of orofacial anomalies in Saudi Arabia.

Keywords: Preliminary report, Cleft deformities, Anomalies, Cleft lip and palate.

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INTRODUCTION

It is now widely recognized that accurate record keeping and data collection is of paramount importance in the running of health services. This is particularly true in the planning and development of service provision and resource allocation, as well as clinical audit and research. Cleft deformities of the face are one of the major congenital anomalies seen in our environment. Prevalence rates for Arab populations, however, have scarcely been studied. In fact, few studies exist that have attempted to report the prevalence of the cleft lip, cleft palate or both among Arab populations.¹⁻³ All available studies have shown a higher prevalence of cleft lip and palate, compared with cleft palate

only⁴⁻⁶ mainly in boys. The left side is affected twice as often as the right side; unilateral cleft lip and palate is more prevalent on the left side and approximately 40% of all cleft infants have associated malformations. Aseer Central Hospital is the major center for cleft surgeries in southern region of Kingdom of Saudi Arabia. Several studies on the management of clefts of the lip and palate have been undertaken, but still there is dearth in data from this major southern center on incidence and prevalence of cleft deformities and associated anomalies which influence management and outcomes. This retrospective study aims to record the pattern of cleft lip/palate and associated anomalies in the hospital.

OBJECTIVES

1. To record the number of patients with orofacial deformities being treated at Maxillofacial Center of Aseer Central Hospital.
2. To determine the pattern of cleft lip and/or cleft palate in Abha of Saudi Arabia from data collected at Aseer Central Hospital, Abha, Saudi Arabia.
3. To lay the foundation for a large population and birth registry based studies.

MATERIALS AND METHODS

The records of all cleft lip and palate patients seen in the hospital from January 2005 to 2011 February were sought and available case notes were reviewed retrospectively. The aim was to gather information from 2005 onward specifically noting the gender, cleft type, presence of an associated syndrome, medical history, where applicable. The cleft deformities were recorded using an anatomically descriptive classification as used in the CARE registration document.⁷

The classification therefore depicted the cleft types as follows:

1. Cleft lip and alveolus
2. Cleft lip and palate
3. Bilateral cleft lip and palate
4. Cleft palate
5. Submucous cleft.

The source of information was from hospital operating lists and plastic surgery department records.

Data sought included the demographic data, types of associated anomalies, completion of procedures among

different types and pattern of follow-up. The detailed description of the associated anomalies was unavailable in many instances and there was no uniform pattern of reporting postoperative results and complications.

RESULTS

Of the 54 patients, 32 (60%) were male and 22 (40%) were female (Table 1 and Graph 1). Isolated cleft palate was most common with around 60% of the total cleft lip/palate patients. In this 56% were females and 44% were males. The second most common was combined cleft lip palate, which accounted for 24% of the total patients of which 70% were males and 30% were females. The least seen was isolated cleft lip accounting to 16% of the total patients seen only in male patients (Table 2 and Graph 2).

Regarding associated anomalies (Table 3 and Graph 3), maximum number (11%) was associated with isolated cleft palate, followed by combined cleft lip and palate around 5%. There were no associated anomalies with isolated cleft lip patients.

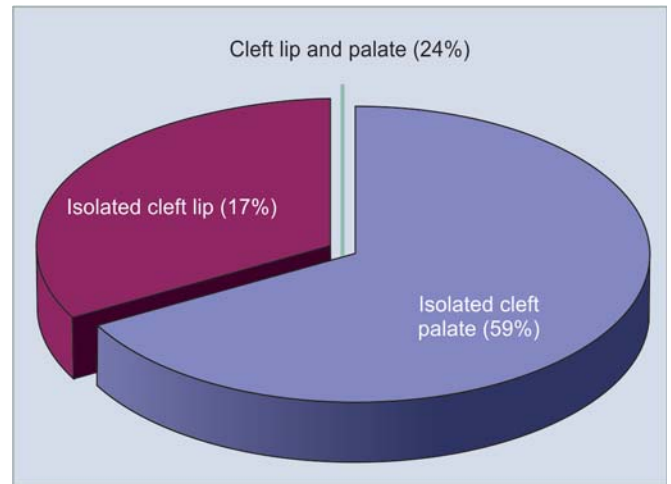
Surgical procedures carried out in the hospital were mainly of cleft palate repair when compared to cleft lip repair (Table 4).

DISCUSSION

The aim of this retrospective study was to report on the pattern of orofacial clefts in Saudi Arabia, based on a review of hospital records at Aseer Central Hospital in Abha. Retrospective studies are usually small, based on clinic

Table 2: Distribution of patients based on cleft pattern

Cleft type	Frequency in male	Frequency in female	Percentage
Isolated cleft palate	14	18	59
Isolated cleft lip	9	0	17
Cleft lip and palate	9	4	24
Total	32	22	100



Graph 2: Distribution of patients based on cleft pattern

records, subject to underreporting, and may suffer from multiple sources of ascertainment bias (e.g. socioeconomic factors).

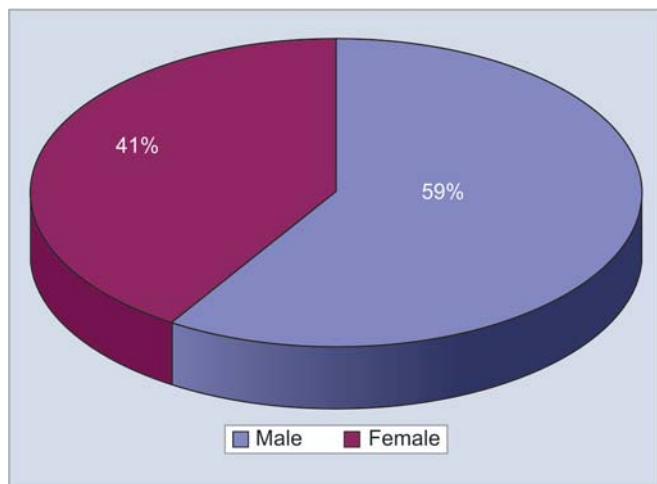
Reports on incidence of associated anomalies vary widely in literature, and have been associated with the manner of data collection.⁸

While cleft lip deformity is obvious and a social stigma, cleft palate is less apparent but associated with more functional problems of regurgitation, speech, recurrent ear and upper airway infections and hearing problems. Esthetic considerations affect the patient’s social acceptance early in life, and speech problems affect the ability to obtain prestigious jobs as adults.⁹ The functional goals of cleft palate surgery are to facilitate normal speech and hearing without significantly affecting the facial growth of the child. Surgical restoration of all components of an abnormal velopharyngeal mechanism at an early age increases the patient’s chances of developing normal speech and hearing.¹⁰

This investigation was undertaken to assess the children born with orofacial clefts, as well as reported patients to Aseer Central Hospital, Abha, Saudi Arabia. Review of the literature revealed very few investigations have been reported regarding the prevalence rate of oral clefts in Arab populations.^{1,2} In the study of Srivastava and Bang (1990) and Al-Bustan et al (2002), the sample was derived from a mixture of native Kuwaitis and expatriate workers of various Arabic nationalities with different socioeconomic and environmental backgrounds.

Table 1: Distribution of oral cleft patients based on gender

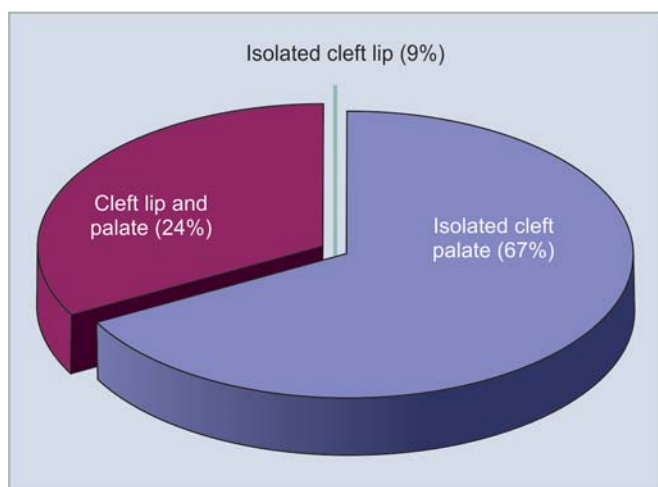
Gender	Frequency	Percentage
Male	32	59
Female	22	41
Total	54	100



Graph 1: Distribution of oral clefts based on gender

Table 3: Distribution of oral cleft patients with associated anomalies

Cleft type	Associated anomalies	Frequency in male	Frequency in female	Percentage
Isolated cleft palate	Pierre Robin syndrome	6	0	67
Cleft lip and palate	Microphthalmia	1	2	33
Isolated cleft lip	–	0	0	0
Total	–	7	2	100

**Graph 3:** Distribution of oral cleft patients with associated anomalies**Table 4:** Surgical procedures done to repair oral clefts

Years	Frequency of cleft lip repair	Frequency of cleft palate repair	Percentage
2005	03	02	09
2008	04	14	30
2009	04	11	26
2010	04	11	26
2011 (till Feb)	03	02	09
Total	18	40	100

Although the exact number is not available, Saudi Arabia has a high rate of consanguineous marriages. Similar marriage customs are common among Arabic populations, with a reported rate of more than 50% of marriages being consanguineous (El-Hazmi et al 1995).¹¹ The present study did not attempt to investigate the effect of close marriages on the occurrence of orofacial cleft in Saudi Arabia. The reported prevalence rate, however, will provide baseline data to investigate this effect.

Most previous studies recognized the limitations of such a retrospective investigation.^{4,12,13} The absence of a compulsory registration system for children born with oral clefts, however, highlights the difficulty in obtaining such data from other than the hospital records. The source of data for the present study was mainly from information recorded in the case notes. The main concern for utilizing the hospital records is a bias in the reported prevalence rate because of the possibility of underreporting the

prevalence.^{13,14} Every attempt was made to eliminate any source of inaccuracies.

The results of the present investigation are in close agreement with the reported prevalence rate for the mixed Arab population in Kuwait (Srivastava and Bang, 1990). Both investigations have found a similar pattern of orofacial clefting and prevalence rates to that reported in white Caucasians.

The difficulty of direct comparison of the findings of the present investigation with the reported prevalence in different races has been addressed in several studies and reviews (Vanderas 1987, Sayetta et al 1989). Differences in the sample selection; data source; inclusion of live, stillbirths or abortions; and classification and definition of the clefts types have been suggested as contributing factors.¹⁵

The results presented here for the 6 years and 1 month period showed minor fluctuations from year to year with no suggestion of a consistent trend toward a change in the prevalence rates (Table 1 and Graph 1). Previous studies have shown a similar trend.^{16,17} Few investigations have found that the prevalence of the cleft lip, cleft palate or both rises during the period studied.^{8,18,19}

The distribution of the types of orofacial clefts was similar to the reported orofacial cleft types distributions among other population groups.^{20,21} The percentage of isolated cleft palate (60%), however, was higher than the value reported in white Caucasians (30 vs 56%), and mixed Arab population in Kuwait (27.8 and 22.4%) and in the Japanese population (18.7 and 21%).²² The results reported here for the number of subjects with cleft with associated anomalies and syndromes indicate only the number and percentage of the subjects with cleft with the associated anomalies, with no attempt to identify these anomalies. Because all the information with regard to any associated anomalies were obtained from hospital records, with limited access to a geneticist in some of the cases, it would appear that the exact diagnosis and manifestation associated with some syndromes might have been missed. The results reported here are thus conservative estimates.

The overall percentage of subjects with cleft with associated major anomalies or syndromes was 16% (Table 2 and Graph 2). The results are in a close agreement with that reported in the Arab population in Kuwait (23.4%), Japanese

population (16.2%) and Argentinian population (23%). Results however, vary markedly from that reported for Caucasians. Fitzpatrick et al (1994) found the percentages of associated anomalies to be as high as 39.5% for the cleft palate group and 25.6% for the cleft lip and palate group. Similar percentages were reported in other studies (Saxen and Lahti 1974, Derijcke et al 1996). However in another study in Saudi Arabia, they reported that 24.4% had only cranial anomalies, 58.4% had only facial anomalies and 17.2% had both of these conditions.²³

The deformities of cleft lip and palate create a challenge for the health care professional involved in their management. Many recommendations were made to ensure that these children receive the most cost-effective and appropriate treatment in a coordinated manner within the settings of a multidisciplinary team. The need for an accurate database for cleft registrations as well as systematic record keeping is essential. This is of particular importance when limited resources exist for planning and developing multidisciplinary teams for treating children with cleft lip, cleft palate, or both.²⁴ The findings of the present investigation provide the number of children with oral clefts being reported to Aseer Central Hospital and thus shall assist in establishing the basis for a database and organizing the treatment for patients with cleft lip and palate in Abha, Saudi Arabia.

CONCLUSION

Isolated cleft palate was more common than the other variants of orofacial clefts. However, pattern of cleft observed in the current study was similar to those reported in the literature for Arab population.

Since this was a hospital-based retrospective study, it was not possible to estimate the true prevalence of orofacial clefts and their associated congenital anomalies in Saudi Arabia at time. Further, large population and birth registry based studies are needed to obtain more precise results regarding both the prevalence and frequency of associated anomalies for orofacial clefts in Saudi Arabia.

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Department of Maxillofacial Surgery, Pediatrics and Medical records. Aseer Central Hospital, Abha, Saudi Arabia.

REFERENCES

1. Srivastava, Bang RL. Facial clefting in Kuwait and England: A comparative study. *Br J Plast Surg* 1990;43:457-62.
2. Al-Bustan SA, El-Zwahr MM, Al-Adsani AM, Bang RL, Ghunaim I, Maher BS, et al. Epidemiological and genetic study of 121 cases of oral clefts in Kuwait. *Orthod Craniofac Res* 2002;5:54-60.
3. Al Omari F, Al-Omari IK. Cleft lip and palate in Jordan: Birth prevalence rate. *Cleft Palate Craniofac J* 2004 Nov;41(6):609-12.
4. Vanderas AP. Incidence of cleft lip, cleft palate, cleft lip and palate among races: A review. *Cleft Palate J* 1987;24:216-25.
5. Coupland MA, Coupland AI. Seasonality, incidence and sex distribution of cleft lip and palate births in Trent Region, 1973-1982. *Cleft Palate J* 1988;25:33-37.
6. Fitzpatrick DR, Raine PA, Boorman JG. Facial clefts in the west of Scotland in the period 1980-1984: Epidemiology and genetic diagnoses. *J Med Genet* 1994;31:126-29.
7. Clinical Standard Advisory Group Report in CARE Newsletter. March 1998.
8. Slavkin HC. Incidence of cleft lips, palates rising. *J Am Dent Assoc* 1992;123:61-65.
9. Greene JC, Vermilion JR, Hay S, Gibbens SF, Kerschbaum S. Epidemiologic study of cleft lip and cleft palate in four states. *J Am Dent Assoc* 1964;68:387-404.
10. Dorf D, Curtin JW. Early cleft palate repair and speech outcome. *Plast Reconstr Surg* 1982;70:75-79.
11. El-Hazmi, Al-Swailem AR, Warsy AS, Al-Swailem AM, Sulaimani R, Al-Meshari AA. Consanguinity among the Saudi Arabian population. *J Med Genet* 1995;32:623-26.
12. Derijcke, Eerens AA, Carels C. The incidence of oral clefts: A review. *Br J Oral Maxillofac Surg* 1996;34:488-94.
13. Sayetta RB, Weinrich MC, Coston GN. Incidence and prevalence of cleft lip and palate: What we think we know. *Cleft Palate J* 1989;26:242-47.
14. Dai L, Zhu J, Mao M, Li Y, Deng Y, Wang Y, et al. Time trends in oral clefts in Chinese newborns: Data from the Chinese National Birth Defects Monitoring Network. *Birth Defects Res A Clin Mol Teratol* 2010 Jan;88(1):41-47.
15. Martelli H Jr, Porto LV, Martelli DR, Bonan PR, Freitas AB, Della Coletta R. Prevalence of nonsyndromic oral clefts in a reference hospital in the state of Minas Gerais, Brazil, between 2000-2005. *Braz Oral Res* 2007 Oct-Dec;21(4):314-17.
16. Gregg T, Boyd D, Richardson A. The incidence of cleft lip and palate in Northern Ireland from 1980-1990. *Br J Orthod* 1994;21:387-92.
17. Bellis TH, Wohlgemuth B. The incidence of cleft lip and palate deformities in the south-east of Scotland (1971-1990). *Br J Orthod* 1999;26:121-25.
18. Fogh Andersen P. Incidence of cleft lip and palate: Constant or increasing? *Acta Chir Scand* 1961;122:106-11.
19. Rintala A, Stegars T. Increasing incidence of clefts in Finland: Reliability of hospital records and central register of congenital malformations. *Scand J Plast Reconstr Surg* 1982;16:35-40.
20. Iregbulem LM. The incidence of cleft lip and palate in Nigeria. *Cleft Palate J* 1982;19:201-05.
21. Rodrigues K, Sena MF, Roncalli AG. Prevalence of orofacial clefts and social factors in Brazil. *Braz Oral Res* 2009 Jan-Mar;23(1):38-42.
22. Natsume N, Kawai T, Kohama G, Teshima T, Kochi S, Ohashi Y, et al. Incidence of cleft lip or palate in 303738 Japanese babies born between 1994 and 1995. *Br J Oral Maxillofac Surg* 2000 Dec;38(6):605-07.
23. Aziza A, Kandasamy R, Shazia S. Pattern of craniofacial anomalies seen in a tertiary care hospital in Saudi Arabia. *Ann Saudi Med* 2011 Sep-Oct;31(5):488-93.
24. Shaw WC, Asher-McDade C, Brattström V, Dahl E, McWilliam J, Mølsted K, et al. A six-center international study of treatment outcome in patients with clefts of the lip and palate: Part I. Principles and study design. *Cleft Palate Craniofac J* 1992 Sep;29(5):393-97.

ABOUT THE AUTHORS**Rafi A Togoo (Corresponding Author)**

Associate Professor, Department of Pedodontics, College of Dentistry
King Khalid University, PO Box 3263, Abha-61471, Saudi Arabia
Phone: 966-7-2418046, e-mail: ratogo@kku.edu.sa

Syed Mohammad Yaseen

Assistant Professor, Department of Pedodontics, College of Dentistry
King Khalid University, Abha, Saudi Arabia

Zakirulla Meer

Assistant Professor, Department of Pedodontics, College of Dentistry
King Khalid University, Abha, Saudi Arabia

Ahmad AIMohy

Intern, Department of Pedodontics, College of Dentistry, King Khalid
University, Abha, Saudi Arabia

Jaber AIQahtani

Intern, Department of Pedodontics, College of Dentistry, King Khalid
University, Abha, Saudi Arabia