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ORIGINAL ARTICLE

Birth prevalence of orofacial cleft in a tertiary hospital in Riyadh, Saudi Arabia: A retrospective audit

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KEYWORD

Craniofacial Anomalies;
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Abstract *Aim:* The present study aimed to calculate the birth prevalence of orofacial cleft patients in King Abdulaziz Medical City (KAMC) Tertiary Care Hospital in Riyadh City, KSA.

Material and methods: The data utilized in this retrospective study were obtained from the birth data registry at the hospital covering the period between January 2014 and December 2018. The data collected for the orofacial cleft patients pertained to the birth year, type of cleft, gender, and associated syndromes.

Results: The total number of documented orofacial cleft cases born between 2014 and 2018 was 78, with a birth prevalence of 1.8 per 1000 live births and no gender bias. Cleft palate (CP) represented the most common type at 38.5% (n = 30), followed by cleft lip at 26.9% (n = 21) and unilateral cleft lip and palate at 24% (n = 19). The least common type was facial cleft, at 3.4% (n = 6). Syndrome association was seen in 15.4% (n = 12) of the cases.

Conclusion: The birth prevalence of orofacial clefts in KAMC Riyadh city is in accordance with the worldwide ratio and similar to the birth prevalence reported in the Middle East area. CP showed more prevalence than the other orofacial cleft types, and the association with syndromes was significantly low.

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1. Introduction

Orofacial clefts, particularly cleft lip and palate, are the most common congenital deformities among newborns. Orofacial clefts represent 65% of all head and neck anomalies (Gorlin et al., 2001). Both environmental and genetic factors con-

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tribute to the etiology of orofacial clefts. Although the cause of orofacial clefts is complex, there is no single risk factor that has been identified in the literature (Mossey, 2007).

The birth prevalence of orofacial clefts varies between different ethnic groups and geographic areas, with the highest prevalence in the Asian population and the lowest in Africa. The prevalence of orofacial clefts has been reported to vary from 0.19 to 2.69 per 1000 live births, with the most accepted incidence rate worldwide being 1 in every 700 live births (Mossey, 2007).

In Riyadh, there have been few reports on the prevalence of orofacial clefts (Aljohar et al., 2008; Kumar et al., 1991). There is no available data regarding the prevalence of orofacial clefts from King Abdulaziz Medical City (KAMC), Riyadh, KSA, which is a 1025-bed capacity facility that provides health care services to the National Guard forces and their dependents, as well as civilian employees and their dependents.

The lack of a birth defect registry at KAMC means the exact count of orofacial cleft patients is unknown. The oral cleft and craniofacial anomaly department is newly established at the hospital, and knowledge of the birth prevalence of orofacial clefts will facilitate a better understanding of the health impact of these defects and help improve services for better patient care.

The null hypotheses of the study were: (1) There is no difference in gender preference regarding the prevalence of congenital cleft defects, (2) There is no difference in laterality regarding the prevalence of congenital cleft defects, (3) There is no difference in the type of cleft regarding the prevalence of congenital cleft defects, and (4) There are no differences in the associated syndromes' status regarding the prevalence of congenital cleft defects.

This study aimed to estimate the birth prevalence of orofacial clefts among live births at KAMC, Riyadh city, KSA.

2. Materials and methods

This retrospective observational study was conducted at KAMC. Riyadh is the largest city in Saudi Arabia and contains about 12% of the Saudi population, an estimated 4 million people (Census, 2020). Unfortunately, there is no national birth defect registry system in Saudi Arabia. At the KAMC, an initial electronic registry system has been fully active since 2014, which records information such as the patient's name, sex, age, and type of orofacial cleft, as well as the treatment interventions and outcomes. The medical records of orofacial cleft patients from January 2014 to December 2018 were extracted from this registry system, and information pertaining to the type of cleft, gender, year of birth, and syndrome affiliation were tabulated.

For the purpose of the study and due to the lack of standardized records in the preliminary registry system at KAMC, orofacial clefts were classified using Davis and Ritchie's system (1922) as cleft lip (CL; right or left, unilateral or bilateral), unilateral cleft lip and palate (UCLP; left or right), cleft palate (CP; soft, hard, or both), bilateral cleft lip and palate (BCLP; left or right), and facial (FC).

Table 1 Demographic Distribution of Study Population and Birth Prevalence per 1000 among the Group.

Total		Total number (%)	Prevalence per 1000 live births
Gender	Male	39 (50%)	0.14
	Female	39 (50%)	0.27
Cleft type	CL	21 (26.9%)	0.50
	CP	30 (38.5%)	0.71
	UCLP	19 (24.4%)	0.45
	BCLP	8 (10.3%)	0.19
	FC	6 (3.4%)	0.14
Medical status	Syndromic	12 (15%)	0.28
	Non-Syndromic	66 (84.6%)	1.57
Total number		78	1.8

3. Statistical analysis

Descriptive statistics for each type of cleft were tabulated, and birth prevalence per 1000 live births was calculated using the total number of live births with orofacial clefts born in the hospital and the total number of births. One sample was used to examine the influence of gender, type of cleft, and side of cleft on the birth prevalence of orofacial clefts. A one-sample binomial test was used to determine the influence of underlying medical condition and gender on the incidence of clefts. The trend of birth prevalence across the years was measured using the Jonckheere-Terpstra trend analysis. All statistical analyses were performed using SPSS ver. 25 (IBM-SPSS, Armonk, NY, USA). Statistical significance was set at $p < 0.05$ for all tests.

4. Results

The sample comprised 78 cases registered in the KAMC hospital database between January 2014 and December 2018. The overall incidence of cleft is summarized in Table 1. There

Table 2 Acceptance or rejection of the null hypotheses.

No.	Null Hypothesis	Test	Sig.	Decision
1	There is no difference in gender preference regarding the prevalence of congenital cleft defects.	One-sample binomial Test	1.000	Retain the null hypothesis
2	There is no difference in laterality regarding the prevalence of congenital cleft defects.	One-sample binomial Test	0.082	Retain the null hypothesis
3	There is no difference in the type of cleft regarding the prevalence of congenital cleft defects.	One-sample binomial Test	0.006	Reject the null hypothesis
4	There are no differences in the associated syndromes' status regarding the prevalence of congenital cleft defects.	One-sample binomial Test	0.000	Reject the null hypothesis

Table 3 Birth prevalence across the years.

Description	Test	Sig.
The distribution of prevalence is the same across categories of year	Independent-samples Jonckheere-Terpstra test for ordered alternatives	0.327

Asymptomatic significances are displayed. The significance level is 0.05.

was an equal distribution of males (n = 39) and females (n = 39) in the sample. Isolated cleft palate (CP) (n = 30) accounted for the greatest number of cases, while facial clefts (FC) (n = 6) accounted for the least (p = 0.006). There were significantly more cases of non-syndromic clefts (84.6%) than syndromic (15%) (p < 0.001). Although there were more unilateral clefts on the left side, the differences were not statistically significant (p = 0.082). The significance of gender, laterality, type, and association with syndromes is depicted in Table 2.

When the trend of birth prevalence was measured across years, it varied from as low as 1 per 1000 live births in 2018 to 2.4 per 1000 live births in 2014 (Table 3). The Jonckheere-Terpstra trend analysis showed that there was no significance in the trend of birth prevalence across the years (p = 0.327).

5. Discussion

Orofacial clefts are associated with a variety of complications, including poor feeding, hearing loss, low self-esteem, and speech difficulties, which significantly reduce both the patients' and their caregivers' quality of life (Queiroz Herkrath et al., 2015) The lack of or inconsistencies in the literature regarding orofacial clefts creates a challenge in understanding their health impact.

The birth prevalence of orofacial clefts in this study was consistent with the globally reported prevalence of 1 in every 700 live births according to the World Health Organization database (WHO Registry Meeting on Craniofacial Anomalies, 2001; Bauru et al., n.d.) Additionally, our results were consistent with the overall mean prevalence of orofacial clefts in the Middle East area including previously published data in Riyadh city, which was documented as 1.25 per 1000 live births (Sabbagh et al., 2012).

In Saudi Arabia, there are few published studies regarding the incidence of orofacial clefts, with high variation, ranging from 0.3 per 1000 live births in Riyadh (Aljohar et al., 2008) to 2.19 per 1000 live births in Al-Qassim (Borkar et al., 1993). This reported variation could be attributed to the lack of a universal registration system within the country.

In this study, although there was a decrease in the birth prevalence of orofacial clefts, it was not statistically significant. The decrease in the orofacial cleft prevalence may be related to the application of the folic acid consumption policy and prenatal counseling in KAMC Hospital. This fact should be correlated with parental data and should become the basis for enforcing new premarital screening test laws within the country.

No difference in gender was detected in this study, and the birth prevalence of orofacial clefts showed a 1:1 male to female ratio, thus retaining the first null hypothesis. This result differs from certain studies, which showed that males were more commonly affected (Impellizzeri et al., 2019), and others that reported a higher prevalence in females (Suleiman et al., 2005).

In the study, although there was no significance in the cleft laterality, the majority of orofacial clefts occurred on the left side, which was in line with several previously published studies (Al Omari and Al-Omari, 2004; Aljohar et al., 2008; Freitas et al., 2004). This fact is related to the slow development of the left facial artery compared to the right (Hirayama, 1971).

In this study, CP was the most common type of orofacial cleft when compared to CL and CLP, thereby rejecting the third null hypothesis, which is inconsistent with what is reported in the literature, wherein different predominance of cleft patterns were recorded (Mossey and Model, 2012). This finding may related to the fact that KAMC is a tertiary hospital in Riyadh city where only illegible individual is treated. The greater birth prevalence of orofacial clefts in the study presented as isolated congenital defects, which was statistically significant when compared to orofacial clefts associated with syndromes. Similar results have been reported in the literature (Al Omari and Al-Omari, 2004). Nevertheless, syndromes result from genetic abnormalities, and the genetic etiology of orofacial clefts in the literature strongly associates them with chromosomal abnormalities and syndromes (Agbenorku, 2013).

The registry system exhibited some limitations, such as unavailable demographic data, including consanguinity, which were obtained from manual files, and lack of classification in a few cases. This had no real impact on the overall results. In the future, a standardized computerized register system with more formative data related to orofacial cleft patients in KAMC cleft centers is needed.

6. Conclusion

The overall prevalence of orofacial clefts at KAMC, Riyadh, Saudi Arabia follows the global pattern of orofacial clefts as well as those of the Middle East region. Isolated CP was more common than the other forms of orofacial clefts. It was also seen that syndromes' association with orofacial clefts was significantly low compared to isolated cleft within the sample.

Authorship statement

Dr. WA and SA designed and conducted the study including data collection and data analysis. Dr. ShA and Miss SA prepared the manuscript draft with essential input from Dr WA and SA. All authors approved the final manuscript.

CRediT authorship contribution statement

Wasmiya A. AlHayyan: Conceptualization, Methodology, Formal analysis, Writing – original draft. **Samar Al Hayek:** Conceptualization, Writing – review & editing, Supervision, Project administration. **Sara S. AlOtabi:** Data curation, Validation. **Shahad A. AlGhanim:** Resources, Data curation, Software.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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