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The registry of congenital cleft lip and cleft palate in Iran: Three-years pilot results and learned lessons

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ABSTRACT

Background: Cleft lip and cleft palate are the most common maxillofacial congenital defects leading to facial deformity and complications in nutrition, respiration, and social interaction. The current study aims to report early findings of the pilot registry for congenital cleft lip and cleft palate in Iran and discuss limitations and further goals to collect and implicate information of cleft newborn patients.

Materials and Methods: A 3-year hospital-based multicenter prospective cohort study of the congenital cleft lip and cleft palate registry was performed in various hospitals in Iran from 2018 to 2020. Newborns with oral clefts were included, and the type of cleft, location, geographical place, and gender of each patient were recorded. The descriptive statistics were reported as means and percentages analyzed with fisher exact test using SPSS software ver. 22 (IBM, Chicago, US). The P-value<0.05 considered as significant.

Results: From 336 registered newborns with cleft lip and cleft palate, 40.1% were females, while 59.9% were males. The prevalence of lip cleft was 31.5%, while 17.6% have palate cleft. Also, 50.9% suffer from both cleft lip and palate simultaneously (P-value<0.05). The frequent location of clefts were bilateral cleft lip and cleft palate with 50% of the cases (P-value<0.05). Also, 28.2% and 21.8% had right-side and left side unilateral cleft lip and cleft palate, respectively. Tabriz, Mashhad, Tehran were the most frequent location of clefts.

Conclusion: Despite the primary statistics and trends presented in this study, the data quality could improve through some limited modifications. Also, the provinces with a high prevalence of congenital deformities need critical attention and provide adequate healthcare.

Keywords: Cleft lip; Cleft palate; Congenital deformity; Epidemiology.

Introduction

The newborn's abnormal physical structure, performance, and metabolism presented at birth are congenital anomalies. The prevalence of these anomalies varies worldwide [1]. Cleft lip (CL) and cleft palate (CP) are the most common congenital disorders in the oral and maxillofacial region [2].

In addition to facial manifestations, CL and CP may affect speaking, hearing, breathing, and nutritional problems [3]. Patients with these disorders need multi-dimensional surgical and nonsurgical care from birth to adulthood [4]. In addition to a higher rate of mortality and morbidity in these patients [5], their families are usually affected with severe mental and social consequences [6]. Identifying the pattern and prevalence of frequent deformities such as CL and CP improve rehabilitation and treatment planning, leading to better outcomes and reducing the burden of these diseases. The issue of poor healthcare and treatment for CL/CP patients in developing and underdeveloped countries is demonstrated in various studies. Early diagnosis, referring, and treatment play critical roles in reaching better outcomes and prognosis. The process of multi-disciplinary treatment in the early stages is summarized to a one-stage definitive repair for late-presented patients [7-9]. There is no exact estimate of CL/CP patients' number and treatment status in Iran, including performed surgeries, complications, rehabilitation, and outcomes. The current study aims to report early findings of the pilot registry for congenital cleft lip and cleft palate in Iran and discuss limitations and further goals to collect and implicate information of newborn cleft patients.

Materials and Methods

The congenital cleft lip and cleft palate registry was designed as a hospital-based multicenter prospective cohort study. A 3-year pilot phase was performed in various hospitals in Iran from 2018 to 2020. The inclusion criteria presented cleft lip and cleft palate (including lip, palate, or both) in newborns. The type of cleft, location, geographical place, and gender was recorded. All patients were included in the cohort study for further evaluations and follow-ups regarding the treatment process and surgical interventions. The descriptive statistics were reported as means and percentages analyzed with Fisher exact test using SPSS software ver. 22 (IBM, Chicago, US). The P-value<0.05 considered as significant.

Results

From 336 registered newborns with cleft lip and cleft palate, 40.1% were females, while 59.9% were males. The prevalence of lip cleft was 31.5%, while 17.6% have palate cleft. Also, 50.9% suffer from both cleft lip and palate simultaneously. (P-value<0.05) (Figure 1). The frequent location of clefts were bilateral cleft lip and cleft palate with 50% of the cases (P-value<0.05). Also, 28.2% and 21.8% had right-side and left side unilateral cleft lip and cleft palate, respectively. (Figure 2). Analysis of the geographical location of clefts revealed that 7.7% of all registered clefts was located in Tabriz, while Mashhad with 6.6% and Tehran with 6% were the most frequent location of clefts.

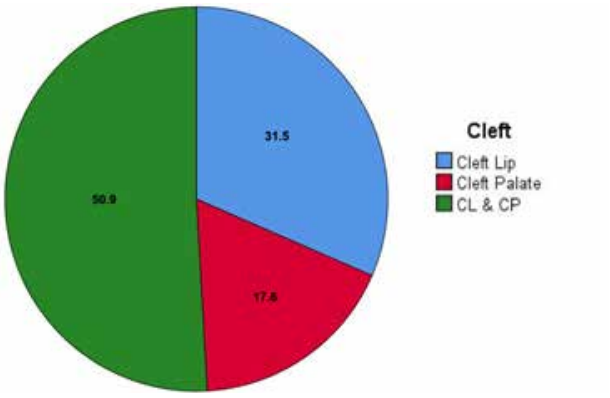

Figure 1. The prevalence of different types of the clefts.

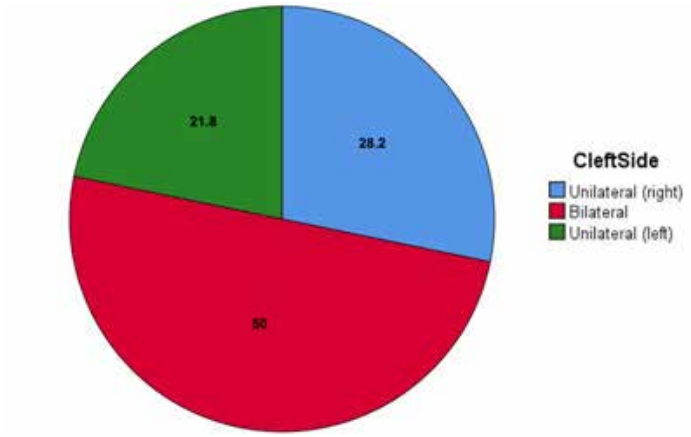

Figure 2. The prevalence of different locations of the clefts.

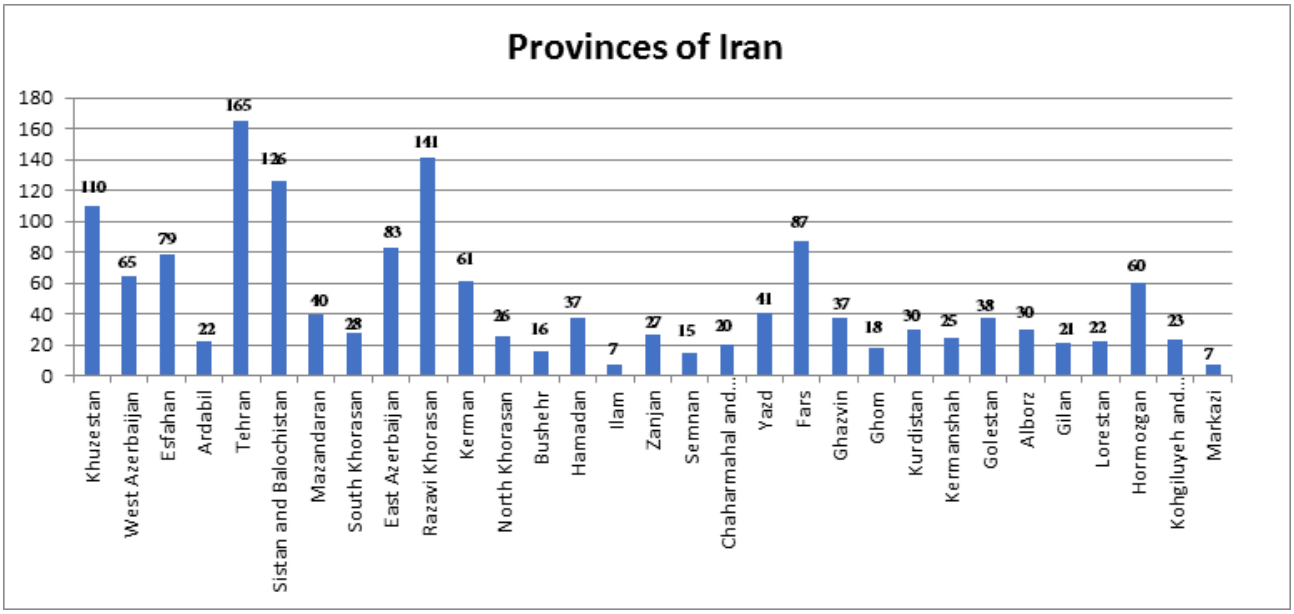

Figure 3. The prevalence of provinces of the clefts.

Discussion

Despite the remarkable decline in the infant mortality rate in developed and underdeveloped countries, congenital abnormalities account as the leading risk factor for an increasing proportion of deaths during the infancy period worldwide [10,11]. Golalipour et al. [12] indicated that the overall prevalence of oral cleft is 0.97 per 1000 live births in Iran. Also, some studies have reported different prevalences of CL and CP in different provinces of Iran. Geographical verities are reported, such as more prevalence of congenital oral defects in the southwest of Iran [13]. Golalipour et al. demonstrated a range of 0.86 to 1.47 per 1000 in different ethnicities in Iran, aligned with the findings of Yassaei et al. [12,14]. In contrast, Sabaq et al. reported a range from 0.3 to 2.4 per 1000 live birth in other Middle-Eastern countries [15]. However, in a hospital-based study in Tehran, Jamilian et al. [16] reported a prevalence of 2.14 per 1000 live births. These findings, alongside current study results, demonstrate a variety of prevalence worldwide and even in provinces of a country. The etiology of this variety suggested being dedicated to the role of race and genetics in their emergence. Namdar et al. reported a 1.2 CL/CP per 1,000 live births with 52.5% and 47.5% of these cases had CL/CP and CL, respectively. Also, they indicated 71.3% of all CL/CP cases occurred unilaterally [17]. In contrast, the result of the present study demonstrated a 31.5% cleft lip while 17.6% have palate cleft. Also, 50.9% suffer from both cleft lip and palate simultaneously. Rajabian et al. [18] reported a prevalence of 0.8 per 1000 live births for cleft lip and palate in the southwest provinces of Iran with 1.25 men to women

ratio. While our findings indicated a high prevalence in the southeast and southwest of Iran, northeast and central regions are the leading geographical locations in occurrences of CL/CP. These are the main limitation of studying the prevalence of congenital defects. Due to migrations and relocations, different ethnicities are located in major big cities, confusing in understanding the role of race and genetics n hospital-based studies. Like other congenital defects. CL/CP are definitive examples of health inequality in the modern world. Without interventional treatments, CL/CP may lead to a high neonatal mortality rate, except with the mildest defect [19]. Due to the inability of suckle in neonates with CL/CP, they died of starvation within a few days. In addition, the psychosocial impact in cleft lip and cleft palate is not easy to define and determine. However, the finding of increased adult suicide rates in Denmark [20] and the high school drop-out rate, unemployment, behavioral issues, episodes of depression, and low self-esteem [21]are indications of the need for this to be very seriously considered in addressing the issue of CL/CP.

A study by Rajabian and Sherkat in 1669 cases in Iran reported that the prevalence of clefts was 1.03 per 1000 births. Cleft lip (without cleft palate) had a higher (34.9%), and cleft palate alone had a significantly lower prevalence (17.4%) than expected [13]. Literature evaluating the status of CL/CP patients in low- and middle-income countries mainly attributed to epidemiology, treatment, and care of individuals, healthcare access, and resource constraints [22-27]. The City of Bauru in Brazil has developed a centre of excellence for the comprehensive management of individuals with CL/CP more than 40 years ago [28]. A review of challenges in CL/CP care in Africa underscored the lack of reliable data on the prevalence of CL/CP because most of the reported studies are hospital-based [29,30]. These are considered limitations of the current CL/CP registry status in Iran, which is mainly based on hospital-gathered data. Also, the referral system for CL/CP is not sufficient and could not provide complete coverage for patients.

The quality of data in a registry could be enhanced by standardizing inclusion criteria, rigorous definitions of data, training, and data gathering software [31]. The demographic variables and information related to diagnosis and treatment procedures of the current study were extracted from HIS regarding the precision and accuracy of data. This indicates that physicians that complete patient files would have a vital role in precise and accurate data collection. Finally, the loss of follow-up is another major issue in studying congenital defects. Due to poor education of families regarding the treatment process, a considerable percentage of these cases remained un-followed.

Conclusion

The pilot phase of the congenital cleft lip and cleft palate registry taught us the limitations of the data gathering system and possible future considerations leading to the improvement of the quality of results. Descriptive epidemiology in the field of cleft lip and cleft palate is an excellent example of how observation of the patterns and trends of the presentation of disease could provide a better understanding of inequalities in healthcare and future aspects that needs further attention.

Conflict of Interest

There is no conflict of interest to declare.

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