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An epidemiological study of patients with congenital cleft palate in Iran

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Abstract

Introduction: Oral clefts are a commonly occurring congenital malformation, and their incidence varies among different racial and ethnic groups. Despite the significance of congenital abnormalities, accurate statistical information about this condition is not readily available. Registry systems can offer valuable information to decision-makers for effectively managing and controlling this condition.

Methods: The current study is a cross-sectional descriptive analysis of registry system data. It analyzed data from 1461 patients with cleft palate over a 15-year period (2008-2023), including various variables and geographical distribution.

Findings: The study found that the ratio of boys to girls with cleft palate is 1.3 to 1. 4.8% of mothers of babies with cleft palate had a history of underlying disease, with hypothyroidism being the most common at 0.9%. In 54% of cases, the parents were related by kinship. Additionally, 58.31% of babies with cleft palate had other abnormalities. The study concludes that the incidence rate of cleft palate varies, influenced by social and racial factors, in different regions of Iran.

Keywords: Epidemiology; Congenital abnormalities; Cleft palate; Oral clefts; Congenital malformation

1. Introduction

Congenital malformations encompass abnormal morphological, behavioral, functional, and chemical defects present at birth that become more apparent as the child ages[1]. According to a recent World Health Organization (WHO) study, congenital anomalies rank seventeenth among the causes of disease burden worldwide [2]. Oral clefts, including cleft lip, cleft palate, and cleft lip and palate, are among the common congenital anomalies and are the second most frequent after bowleg [3]. Cleft lips and palates are classified into different types, such as unilateral or bilateral and complete or incomplete. Patients experience various problems, including functional disorders like speaking, hearing, chewing, swallowing, respiratory issues, facial and nasal deformities, and feeding difficulties [4].

A cleft of the roof of the mouth is called a cleft palate. The palate consists of two parts: the hard palate and the soft palate, both of which can develop clefts. A cleft palate appears when the palates on both sides fail to fuse between the 3rd and 12th weeks of fetal development, resulting in a gap. The severity of the cleft can vary; it can involve only the palate or, in more severe cases, extend to extensive facial clefts. This condition occurs in all countries and varies in incidence among racial and ethnic groups. Epidemiological studies have shown considerable variations in birth prevalence across different countries.

In different studies conducted worldwide, various rates of incidence and prevalence of cleft palate have been reported. For instance, a study in France in 2012 found an incidence of 1.2 per 1,000 births[5]. Additionally, a meta-analysis in Iran in 2019 revealed a prevalence of 1.24 per 1,000 live births for oral clefts[6]. The occurrence of this disease is influenced by several factors worldwide, including medication use, vitamin and folic acid deficiencies, smoking,

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maternal overweight, geographic location, nationality, nutrition, and maternal hormonal disorders [7]. In Iran, a study by Noorollahian and colleagues identified factors such as the use of psychiatric medications, hypoxia, smoking, vitamin and folic acid deficiencies, maternal obesity and overweight, hormonal disorders, hereditary factors, and geographic location as contributors to the disease [8].

Research reports on congenital diseases are lacking in developing countries. In Iran, the Ministry of Health faces challenges due to the need for accurate statistics on the status of patients. In contrast, developed countries have access to such statistics [9]. In many developed countries, congenital defect prevalence is monitored through registry or surveillance systems[10]. Registries are technological infrastructures that provide standardized information about groups of patients with a specific condition. Registry systems can be useful for healthcare providers to uniquely monitor and track their patients, leading to more appropriate and effective treatments for similar groups. Registries can collect complete, accurate, and high-quality data, assisting healthcare providers and policymakers in creating optimal care strategies. They can also offer high-quality and precise reports for better research and decision-making [11,12].

Registry systems are crucial in reducing the mortality and morbidity associated with diseases and medical interventions [13]. Evidence indicates that congenital anomalies significantly cause hospital admissions and medical care among children [14]. Treating these patients imposes significant costs on society overall [15].

One of the significant challenges in the healthcare sector is ensuring adequate financial resources. In Iran, these resources are obtained through various methods such as taxes, social insurance payments, direct payments, and support from charitable organizations. Over 90% of the budget for charitable organizations comes from external sources. Despite the significant role of charitable organizations in the healthcare sector, they have been neglected in academic research and studies[16]. Therefore, this study was conducted to examine patients with cleft palates who are covered by the Charity Foundation for Supporting Children with Congenital Malformation (MOHKAM).

2. Methods

This study is a descriptive cross-sectional analysis of data collected from patients with cleft palate over 15 years, from 2008 to 2023, in 31 provinces in Iran, covered by MOHKAM. Data were extracted from the MOHKAM charity registry system, the Hospital Information System (HIS), and medical records. The variables examined included age, gender, pregnancy method, history of maternal disease before pregnancy, exposure to chemicals, history of cosmetic use, parental kinship, associated diseases, and geographical distribution. Cases of cleft lip accompanied by cleft palate were considered as having simultaneous defects.

The study included all patients whose medical expenses were covered by the charity foundation. These patients represented all age groups, from birth to young adulthood, and had received treatment. Out of the 8,241 patients with congenital anomalies covered by the foundation, 1,461 cases of cleft palate, all of whom had undergone reconstructive surgery, were selected for the study. Data analysis was carried out using R software. The results were then summarized and presented in tables and charts.

3. Results

In the present study, we found that of 1,461 patients with congenital cleft palate, 820 were male (56.1%), and 641 were female (43.9%). 91.8% of the mothers did not use cosmetics during pregnancy (1,341 out of 1,461). In 6.8% of the cases (100 out of 1,461), the parent's place of residence was exposed to power towers. 4.8% (70 out of 1,461) of the mothers had a history of illness before pregnancy, and in 4.1% of the cases (60 out of 1,461), the mothers reported medication use during pregnancy. 90.5% of the mothers (1,322 out of 1,461) did not have a history of chemical exposure during pregnancy.

Of 1,461 cases, 881 (60.3%) were delivered naturally, and 580 (39.7%) were delivered via cesarean section. 1,452 cases (99.4%) were conceived naturally, while 9 cases (0.6%) were conceived through IVF. Additionally, 672 cases (46%) involved kinship parents (see Table 1).

Table 2 presents the history of illnesses before pregnancy. Among the mothers of infants with cleft palate, 70 (4.79%) had accompanying conditions during pregnancy. These conditions included hypothyroidism, diabetes, anemia, neurological disorders, hypertension, kidney stones, seizures, asthma, migraines, depression, minor thalassemia, epilepsy, and liver cysts, listed in order of prevalence. Additionally, 852 cases (58.31%) of infants with cleft palate had

other abnormalities besides the cleft palate. Among these, 755 infants (51.7%) had both cleft lip and cleft palate (Table 3).

The study examined the ages of patients with cleft palate and their mothers at the time of referral for reconstructive surgery. The results are presented in Tables 4 and 5. The findings show that the average age of mothers was 28.8 years, with the highest frequency in the age range of 25 to 29 years. Likewise, the average age of patients with cleft palate at the time of referral was 10.2 years, with the highest frequency observed in newborns to 5-year-olds (Tables 4 and 5).

From 2008 to 2018, the provinces of Tehran, Khorasan Razavi, and Alborz had the highest number of patients with congenital cleft palate. From 2018 to 2023, the provinces with the highest number of cases were Tehran, Khorasan Razavi, and Sistan and Baluchestan (Table 6).

Table 1 Characteristics of patients with congenital cleft palate

variables		Frequency	Percentage
Sex	Male	820	56.1%
	Female	641	43.9%
Examined Variables	Cosmetic consumption during pregnancy	120	8.2%
	Proximity of the location to the power tower or energy sources	100	6.8%
	History of disease before pregnancy	70	4.8%
	Drug intake during pregnancy	60	4.1%
	Chemical consumption during pregnancy	139	9.5%
Type of delivery	Normal	881	60.3%
	Cesarean	580	39.7%
Method of pregnancy	Normal	1452	99.4%
	IVF	9	0.6%
Parental Kinship	Related	672	46%
	Unrelated	789	54%

Table 2 Distribution of Underlying Diseases among Mothers of Congenital Cleft Palate Patients

Disease	Frequency	Percentage within Diseases Subgroup (n = 70)	Percentage of Total Population (n = 1461)
Hypothyroidism	13	18.60%	0.90%
Diabetes	12	17.10%	0.80%
Anemia	11	15.40%	0.80%
Neurology and Psychiatry	8	11.40%	0.50%
Blood Pressure	7	10.00%	0.50%
Kidney Stone	6	8.60%	0.40%
Convulsions	4	5.70%	0.30%
Asthma	3	4.30%	0.20%
Migraine	2	2.90%	0.20%

Depression	1	1.40%	0.10%
Thalassemia Minor	1	1.40%	0.10%
Epilepsy	1	1.40%	0.10%
Liver Cyst	1	1.40%	0.10%
Total	70	100%	4.80%

Table 3 Distribution of Congenital Comorbidity In Congenital Cleft Palate Patients

Comorbidity	**Frequency**	**Percentage within Comorbidity Group (n = 852)**	**Percentage of Total Population (n = 1461)**
Cleft lip	755	88.60%	51.68%
Congenital heart defects	13	1.50%	0.89%
Clubfoot	8	0.90%	0.55%
Imperforate anus	8	0.90%	0.55%
Esophageal atresia	7	0.80%	0.48%
Hirschsprung's disease	7	0.80%	0.48%
Ventricular septal defect	6	0.70%	0.41%
Growth hormone deficiency	6	0.70%	0.41%
Choledochal cyst	6	0.70%	0.41%
Hydrocephalus	6	0.70%	0.41%
Sexual ambiguity	4	0.50%	0.27%
Neurogenic bladder	4	0.50%	0.27%
Hypospadias	4	1%	0.27%
Stool incontinence	3	0.40%	0.21%
Congenital pelvic dislocation	3	0.40%	0.21%
Omphalocele	2	0.20%	0.14%
Pierre Robin Syndrome	2	0.20%	0.14%
Cataract	2	0.20%	0.14%
Meningocele-myelomeningocele	2	0.20%	0.14%
Nose deviation	1	0.10%	0.07%
Encephalocele	1	0.10%	0.07%
Heart and lung disease	1	0.10%	0.07%
Fallot Tetralogy	1	0.10%	0.07%
Congenital diabetes	1	0.10%	0.07%
Total	852	100%	58.32%

Table 4 Age Distribution of Patients with Cleft Palate

Age (year)	Frequency	Percentage
0 - 5	476	32.60%
6 - 10	363	24.80%
11 - 15	299	20.50%
16 - 20	140	9.60%
21 - 25	90	6.20%
> 25	93	6.40%
Total	1461	100%

Table 5 Age Distribution of Mothers of Cleft Palate Patients

Age	Frequency	Percentage
< 20	100	6.80%
20-24	269	18.40%
25-29	587	40.20%
30-34	255	17.50%
35-39	151	10.30%
> 40	99	6.80%
Total	1461	100%

Table 6 Provincial Distribution of Congenital Cleft Palate Patients

Province	Frequency	Percentage
Alborz	60	4.1%
Ardabil	10	0.7%
Bushehr	17	1.2%
Chaharmahal and Bakhtiari	21	1.4%
East Azarbaijan	12	0.8%
Esfahan	87	6%
Fars	33	2.3%
Ghazvin	18	1.2%
Gilan	13	0.9%
Golestan	27	1.8%
Hamedan	24	1.6%
Hormozgan	78	5.3%
Ilam	19	1.3%

Kerman	51	3.5%
Kermanshah	34	2.3%
Khorasan Razavi	180	12.3%
Khuzestan	91	6.2%
Kohgilooyeh and Boyerahmad	35	2.4%
Kordestan	35	2.4%
Lorestan	73	5%
Markazi	23	1.6%
Mazandaran	24	1.6%
North Khorasan	12	0.8%
Qom	43	2.9%
Semnan	15	1%
Sistan and Baluchestan	105	7.2%
Southern Khorasan	17	1.2%
Tehran	257	17.6%
Western Azerbaijan	19	1.3%
Yazd	15	1%
Zanjan	13	0.9%
Total	1461	100%

4. Discussion

The present study examined a large number of patients with congenital cleft palate across Iran. A total of 1,461 cases of cleft palate were analyzed in terms of factors influencing the occurrence of the condition, associated diseases, geographical distribution in the country, and the age of the mother and patient.

Regarding gender, the results showed that the ratio of boys to girls was 1.3 to 1 among patients with cleft palate. In a study by Jalilevand and colleagues in Iran, the ratio of boys to girls among newborns with clefts in the western and northwestern provinces of Iran was reported to be 1.2 to 1[17]. Mirfazeli and colleagues reported that this ratio was 1.4 to 1 in hospitals in Gorgan[18]. Similar results to those found in studies conducted in Iran have been observed in Argentina, Pakistan, and Croatia[19-21].

In the current study, 46% of the patients' parents had kinship relationships. A study by Ordubazari and colleagues in 1995 reported a 15% rate of kinship in the occurrence of oral clefts[22]. Another study by Sahafian in Mashhad noted a 12% kinship rate for oral clefts[23]. Moreover, Azimi and colleagues reported a 58.1% kinship rate among all patients, with Velo-Cardio-Facial Syndrome being the most commonly associated disease[24].

In this study, 58.3% of infants with cleft palate also had abnormalities in other organs. Among congenital anomalies associated with cleft palate, congenital heart defects were the most common after cleft lip. Similarly, in a study by Habibollahi and colleagues, 50.9% of patients suffered from both cleft lip and palate[25]. In the present study, 4.8% of the mothers of infants with cleft palate had associated diseases, with hypothyroidism, diabetes, anemia, and neurological disorders being the most frequent. In contrast, in the study by Sadri and colleagues, 9.3% of mothers had associated diseases[26].

According to the study by Neves and colleagues, 27.59% of mothers were exposed to chemicals during pregnancy, with 6.1% specifically using cosmetics[27]. However, in the present study, 9.5% of mothers used chemicals, with 8.2% specifically using cosmetics. In the study by Zandi and colleagues, 23.3% of mothers had a history of medication use during pregnancy[28], whereas in the present study, medication use among mothers was 4.1%.

A study in Estonia found that 28.4% of mothers who had children with congenital facial anomalies were under 30 years old[29], which aligns with our study's results. Our study showed that the highest frequency of births with cleft palate (40.2%) occurred in mothers aged 25 to 29. Maternal age has been widely studied as a risk factor for oral clefts and should be considered[30, 31].

In different regions of Iran, the prevalence of congenital cleft palate has been reported to range between 0.485 and 3.73 per 1,000 live births. This variation, especially in Iran, may be due to the presence of different ethnicities and races, differences in dietary habits, environmental factors, or related genetic and cultural factors, as Iran is a country with completely diverse ethnicities and environments. Various studies have reported the impact of these factors on the incidence of oral clefts[6,32].

Due to migration and relocations, different ethnic groups are now located in major cities, making it challenging to understand the role of race and genetics in hospital-based studies. In our study, according to Table 6, over the course of 15 years, the highest number of patients treated came from the provinces of Tehran (17.6%), Khorasan Razavi (12.3%), and Sistan and Baluchestan (7.2%). In another study, the overall prevalence of oral clefts was reported to be 1.24 per 1,000 live births, with the highest rate in Tehran and the lowest in the western and northwestern provinces of the country[33].

It is recommended that cleft lip repair be performed at three months of age[34]. A retrospective study on the epidemiology, clinical aspects, and management of clefts in Burkina Faso reported that more than 60% of children sought treatment after the age of one year[35]. Our study shows that the majority of children (32.6%) were brought to the charity for reconstructive surgery between the ages of 0 and 5.

The first step toward primary prevention is Identifying modifiable risk factors for oral clefts. Such preventive efforts may require changes in maternal lifestyles, improved diet, use of multivitamins and mineral supplements, avoiding certain medications, and raising public awareness of social, occupational, and residential risk factors[35].

5. Conclusion

This study of 1,461 patients with congenital cleft palate in Iran highlights influencing key factors. It reveals that cleft palate is more common in boys and frequently associated with parental kinship, indicating a strong genetic component. A significant number of infants also had other congenital issues, particularly congenital heart defects. Geographic variations in incidence suggest social, racial, and environmental influences, pointing to the need for region-specific public health interventions. The study also links maternal exposure to chemicals and other risk factors during pregnancy with the occurrence of cleft palate, highlighting the importance of preventive measures focused on maternal health. Future initiatives should aim to improve data collection through better registry systems, raise awareness about prevention, and continue research to reduce the incidence and burden of cleft palate in Iran.

Compliance with ethical standards

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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