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Prevalence and Patterns of Congenital Limb Defects in the North of Iran (2007-2011)

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ABSTRACT

Background: Congenital limb defects (CLDs) are the leading cause of disability across the world.CLDsvary depending on the anatomical location, type, and cause of anomaly. The present study aimed to evaluate the prevalence and patterns of CLDs in the North of Iran.

Methods: This descriptive-analytical, hospital-based study was conducted in three hospitals in Gorgan, the capital of Golestan Province in the north of Iran. Samples included 32,895 newborns with CLDs, and stillborn neonates were excluded from the study. Data analysis was performed in SPSS version 16 using Chi-square at the significance level of ≤0.05.

***Results*:** Overall prevalence ofCLDs was three cases per 1,000 live births, and the rate was estimated at 3.17 and 2.82 cases per 1,000 live births in male and female infants, respectively. In addition, the prevalence of upper- and lower-limb anomalies was 1.03 and 1.91 cases per 1,000 live births, respectively. In terms ofethnicity, the prevalence of CLDs among the native Fars, Turkmen, and Sistani populations was 3.86, 2.02, and 3.85 cases per 1,000 live births, respectively. Clubfoot was the most common type of CLDs, and the most prevalent type of the associated malformationswas gastrointestinal anomalies.

Conclusion: According to the results, the prevalence of CLDs in the north of Iran was similar to European countries (2.11-3.18 cases per 1,000 live births), while it was lower compared to the other regions in Iran (5.8 cases per 1,000 live births).

Keywords: Birth defect, Ethnicity, Iran, Limb, Prevalence

Introduction

Limb defects are of a congenital origin and occur when the limbs fail to develop normally during the intrauterine life (1). Congenital limb defects (CLDs)account for the most common birth anomalies in infants after congenital heart anomalies (2).The prevalence of the congenital malformations of the upper limbshas been reported to be one case per 506 live births. CLDs could be isolated, combined with the other limbs or systems, or associated with a specific syndrome.

Environmental and genetic factors have been shown to be involved in the development of the limbs (2).Disruptive events and prenatal exposure to various teratogens are the common causes of CLDs. For instance, thalidomide is considered to be a teratogen in the process of limb development (1). CLDs vary depending on the type, anatomical location, geographical location, ethnicity, and gender and are often associated with lifelong functional handicaps (3).

Reports are variable regarding the rates of CLDs (1, 4-7). Considering the lack of documented data and ethnical variations in our region, the present study aimed to assess the prevalence and patterns of CLDs in the north of Iran during a four-

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year period.

Methods

This descriptive-analytical, hospital-based study was conducted as a population surveillance in three hospitals in Gorgan, the capital of Golestan Provincein the north of Iran during March 2007-2011. The study protocol was approved by the Ethics Committee of Golestan University of Medical Sciences. After the approval of the consent forms by the Ethics Committee, written informed consent was obtained from the mothers of infants to participate in the study.

Subjects were selected fromMasoud Hospital, Falsafi Hospital, and Dezyani Hospital in Gorgan city. Dezyani Hospital is the largest specialized obstetrics and gynecology referral center with 120 beds, and Masoud Hospital (60 beds) and FalsafiHospital (100 beds) are private general hospitals. The three main ethnic groups in the study were Fars, Turkmen, and Sistani populations of Gorgan city. Native Fars is the predominant ethnic group in the region, the Turkmen population emigrated to Gorgan from central Asia more than three centuries ago, and the Sistani population emigrated to Gorgan from the south-east of Iran half a century ago.

All the neonates who were born with CLDsduring the study period were enrolled, and the exclusion criterion was stillbirth. Collected data included the gender of the infant, ethnicity of the parents, and the pattern of CLDs, which were recorded in a questionnaire for each newborn. In each hospital, two staff members (registered nurses) were recruited and trained to review all the birthsand register the limb anomalies. Congenital anomalies were confirmed by a neonatologist, and the classification of the upper- and lower-limb anomalies was performed based on the International Classification of Diseases (ICD-10).

Statistical analysis

Data analysis was performed inSPSSversion 16 using Chi-square, and the P-valueof ≤0.05 was considered statistically significant.

Results

In total, 32,895 infants were born during the study period in Gorgan, 99 of whom had CLDs with the prevalence of three cases per 1,000 live births. The prevalence of CLDs in the male and female neonates was estimated at 3.17 and 2.82cases per 1,000 live births, respectively, and the difference was not considered significant in this regard (P=0.36).

According to the results, clubfoot was the most common type of CLD with the incidence of onecase per 1,000 live births, followed by polydactyly (0.79 per 1,000 live births), meromelia (0.33 per 1,000 live births), achondroplasia (0.33 per 1,000 live births), and congenital hip dislocation (0.30 per 1,000 live births).

Among the studied newborns, 68.7% had additional congenital anomalies. In this regard, the most frequent associated anomalies were gastrointestinal anomalies (28.3%), central nervous system anomalies (20.2%), cardiovascular anomalies (14.1%), and urogenital anomalies (12.1%) (Table 1).

According to the findings, 30.3% of the neonates had upper-limb anomalies, while 49.5%of the CLDs were detected in the lower extremities (Table 2). Moreover, the prevalence of upper- and lower-limb CLDs was determined to be 1.03 and 1.91 cases per 1,000 live births, respectively.

Table 1. Frequency of Associated Anomalies in Anomalous Events; Gorgan, Northern Iran, 2007–2011

Type of CLD	Number of Associated Anomalies	%
Digestive System	28	29.16
Central Nervous System and Neural Tube Defects	20	20.83
Cardiovascular and Respiratory Systems	14	14.58
Urogenital System	12	12.50
Other Anomalies	22	22.92

In terms of ethnicity, the prevalence of CLDs among the native Fars, Turkmen, and Sistani populations was 3.86, 2.02, and 3.85 cases per 1,000 live births, respectively (Table 3). Changes occurring in theCLDs during the study period are depicted in Figure 1. Accordingly, the highest prevalence of CLDs was in 2010.

Discussion

According to the results of the present study, theprevalence of CLDs was 3.00 cases per 1,000 live births in Gorgan, which is higher than the findings of Vasluian et al. (2.11 cases per 1,000 live births) (1).Furthemore, the prevalcne of CLDs in the current reesarch was higher compared to

Table 2. Type and Number of CLDs

CLD	N	%
Type of Upper-Limb Anomalies		
Polydactyly	17	31.50
Syndactyly	6	4.76
Lobster-claw Hand	2	1.58
Meromelia	6	4.76
Other	3	2.40
Type of Lower-Limb Anomalies		
Clubfoot	33	26.20
Syndactyly	1	0.80
CDH	10	8
Meromelia	5	4
Polydactyly	9	7.14
Micromelia	2	1.58
Amputation	2	1.58
Rhizomelia	1	0.80
Type of Upper- and Lower-Limb Anomalies		
Achondrogenesis	1	0.80
Bone Dysplasia	8	6.35
Achondroplasia	11	8.73
DMD	2	1.58
Other	7	5.55

Table 3. Prevalence of CLDs in Terms of Ethnicity

Variable	Population	Delivered Newborns (n)	Newborns with CLD(n)	Rate per 1,000 Live Births
Ethnicity	Fars	13,488	52	3.86
	Turkmen	10,855	22	2.02
	Sistani	4,934	19	3.85

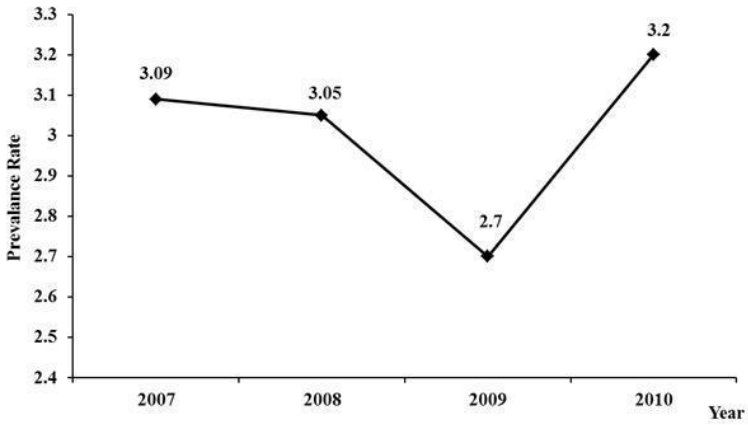

Figure 1. Prevalence Rate of Congenital Limb Anomalies (CLDs) per 1,000 Live Births

the studies by Gerber Ekblom et al.(2.15 cases per 1,000 live births) (4) and Shawky et al. (1.80 cases per 1,000 live births) (5).

In another research by Koskimies (6), the prevalence of upper-limb anomalies was reported to be 0.52 per 1,000 live births. In the EUROCAT report, the prevalence of CLDs was estimated at 3.80 cases per 1,000 live births, which is consistent with the present study (8). On the other hand, the estimated prevalence of CLDs in our study was compared to a research conducted in Ahvaz (south of Iran) (7), which was reported to be 5.89 cases per 1,000 live births (Table 4).

According to the current research, clubfoot and polydactyly were the most common CLDs, while in the study by Vasluian et al. in the Netherlands (1), polydactyly, reduction defects, and syndactyly were reported to be the most common limb abnormalities. Moreover, the findings of Sarrafan et al. in the south of Iran indicated that clubfoot, congenital dislocation of hip, and polydactyly were the most prevalent CLDs, which is in line with our findings(7). In a study inUganda,clubfoot was the most frequent type of CLD in the live

Table 4. Incidence of CLDs in Various Regions of the World

Author(s)	Location	Study Period	Incidence per 1,000 Live Births
Present Study	Gorgan (North of Iran)	2007-2011	3
Shawky et al. (5)	Egypt	1995-2009	1.8
Giele et al. (10) (CULA*)	Western Australia	1980-1990	2
Vasluian et al. (1)	The Netherlands	1981-2010	2.11
Gerber Ekblom et al. (4) (CULA)	Sweden	1997-2007	2.15
Sarkar et al. (11)	Eastern India	2011-2012	0.32
Sarrafan et al. (7)	Ahvaz (South of Iran)	2006-2007	5.8

births (9).

Gastrointestinal, central nervous system, cardiovascular, and urogenital system defects were observed to be the most common associated congenital anomalies in the north of Iran, respectively, which is similar to the results obtained byVasluian et al. in the Netherlands(1). On the other hand, congenital malformations in thecirculatory system, head and neck, and urogenitaland digestive systemshave beenregarded as the most prevalent types of associated anomalies in Sweden (4).

In terms of gender, thefindings in Sweden (4) and Western Australia (10) are in congrunce with the results of the present study, denoting that congenital upper-limb anomalies are more common in male neonates compared to female neonates. In terms of ethnicity, the prevalence of CLDswas found to be lower in the Turkmen population compared to the other ethnic groups in the north of Iran.In contrast with our finding,no significant differences wereobserved between the white and non-white newbornsin terms of the prevalence of CLDs in Western Australia (10).

In the study by Vasluian et al. in the Netherlands(1), upper limbswere more commonly affected by CLDscompared to the lower limbs (lower limb to upper limb ratio: 1:2), which is inconsistent with the results of the current research, as lower limbswere more frequently affected by the CLDs compared to the upper limbs. This discrepancy could be due to the differences in racial/ethnical, geographical, socioeconomic, and nutritional factors.

Conclusion

According to the results, the prevalence rate of CLDs in the north of Iran was similar to European countries (2.11-3.18 cases per 1,000 live births), while it was lower than the other regions in Iran (5.80 cases per 1,000 live births).Further studies are required to determine the exact etiology of CLDs, and our findings could help establish a database for further investigations to determine the causes of limb anomalies in the north of Iran. Moreover, the provided evidence could be used to modify health policies in order to improve the diagnosis and treatment of CLDs.

Limitations of the study

One of the limitations of the current research was disregarding the stillbirths, which might have affected the estimated prevalence rate of CLDs. In addition, our study was a hospital-basedsurvey, which cannot ensure accuracy similar to generalpopulation-based studies.

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Conflicts of interests

None declared.

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