Incidence of Clefts in Israel

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A previous survey (unpublished) on the incidence of congenital cleft lip and/or palate was carried out in 1959. That incidence rate was found to be 1/1209 ± 0.38%. That rate was lower than the usually reported frequency, and it was assumed that the difference between that rate and those rates reported in the literature might have been due to incorrect or incomplete registration of these malformations in our hospitals. It was, therefore, decided to repeat the survey on a larger scale after communicating with the Ministry of Health and giving instructions to the maternity wards for exacting registrations. The purpose of this paper is to define the incidence rates of congenital facial clefts among the Jewish population in Israel and to compare it with the incidence in other countries, as reported in the literature.

Materials and Methods

The records of the maternity wards in six regional hospitals in the different sections in Israel were screened. These hospitals were judged to provide a representative sample of the Jewish children born in Israel; indeed, 95% of all deliveries take place in those hospitals. Children of the different minorities were not included in the present survey, since it is not customary for their mothers to be routinely delivered in these hospitals and thus no valid registration of congenital deformities among such groups was available.

The collected data were submitted to statistical analysis, that is, the incidence rate was estimated on the basis of 1,000 live births. For the statistical analysis, the chi square test was used to test the independence and the normal approximation for the binomial distribution. The level of significance was 5%. The total live births in each hospital were tested for differences with the remaining five hospitals. Differences in number of births between the years 1960, 1961, and 1962, and differences between the sum total of live births in all six hospitals for each of the years were also tested.

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TABLE 1. Incidence of cleft lip and cleft palate per number of live births.

	1960		*1961*		*1962*		*total*	
	N clefts	*N live births*	*N clefts*	*N live births*	*N clefts*	*N live births*	*N clefts*	*N live births*
number of births	9	16,547	6	15,812	11	15,409	26	47,768

TABLE 2. Distribution of clefts according to type and sex for the period 1960–1962.

type of clefts	*M*		*F*	
	N	%	*N*	%
cleft lip	2	7.7	6	23.1
cleft palate	6	23.1	2	7.7
cleft lip, palate	5	19.23	5	19.23
total	13	50.00	13	50.00

Findings

No statistically significant differences were found between the six hospitals, or between the years 1960, 1961, and 1962. On this basis, the combined sample for the three years in the six hospitals was used for the survey (Table 1). During the period 1960 through 1962, the incidence of cleft lip and cleft palate was one in 1,837 live births. The rate per 1,000 was 0.544, with a standard error of ±0.0001.

The distribution of clefts between the sexes showed no significant predilection, nor were there significant differences between the sexes with respect to the incidence of the types of clefts (Table 2).

Data regarding age of mother are presented in Table 3, assuming that the age distribution of the mothers included in this study was comparable to the age distribution of the total population of mothers during the years 1960, 1961, and 1962.[1] A trend for higher incidence of clefts in the youngest age group is apparent. Due to the fact that this trend becomes apparent, based on the above assumption, and due to the small number of cases in each group, no further statistical tests were carried out.

Data regarding birth order were also calculated on the assumption that the distribution by birth rank among the surveyed cleft palate cases was the same as that of all births during 1960, 1961, and 1962. No significant trend was apparent (Table 4).

Multiple malformations accompanying cleft lip and cleft palate were

[1] Figures for the age distribution of mothers for these years were obtained from the Statistical Year Book, 1964.

TABLE 3. Number of clefts by age of mother for the period 1960–1962. (Information about mother's age for one child was not available.)

age of mother	*N*	*rate per 10,000*
20–24	10	6.9
25–29	8	5.5
30–34	5	5.4
35–39	2	4.3
total	25	5.4

TABLE 4. Number of clefts by birth rank. (Information about birth rank for one child was not available.)

birth rank	*N*	*rate per 10,000*
1	7	5.5
2	7	5.8
3	5	6.8
4+	6	3.8
total	25	5.4

TABLE 5. Number of clefts by mother's country of origin. (Information was missing for one child.)

Country of origin	*N*	*rates per 10,000*
Asia	10	6.817
Africa	4	2.881
Europe-America	3	2.825
Israel	8	4.314
total	25	

found in 27% of the cases. As many as six developmental anomalies were registered in one single child. There was an even distribution of multiple anomalies between the sexes.

The number of live births by mother's country of origin was estimated by the frequency distribution of deliveries among these populations in Israel during the period of 1960–1962, and was utilized as denominators in calculating the rates of cases with clefts. From this group estimation, trends are apparent that the incidence of clefts is higher among the populations from Asian countries (Table 5).

In the present survey the incidence of congenital cleft lip and palate was found to be one in 1,837 live births. Compared to the incidence rate reported from other countries in recent years (Table 6), it is

TABLE 6. Frequency of cleft lip and palate in various countries. Portions of this material came from Greene (*5*), with consent of author and editor. For the 1918 study and for 1938 study, according to data given, incidence rate should be .58 and 1.94 per 1,000, respectively.

Year	*Location*	*Source*	*Number of clefts*	*Sample size*	*Clefts per 1,000*	*Population per cleft*
1864	St. Petersburg, Russia	foundling hospital	118	180,000	0.66	1,525
1908	London, England	hospital admissions	39	67,945	.57	1,742
1918–19	United States	army draftees	1,466	2,510,791	.53	1,880
1926	Baltimore, Md.	birth records:	24	28,085	.85	1,170
		White	17	15,565	1.09	915
		Negro	7	12,550	.56	1,793
1931	Lubeck, Germany	population survey	28	34,000	.82	1,214
		birth records	102	102,823	.99	1,008
1934	Holland	babies	16	15,270	1.05	954
1934	Hamburg, Germany	birth records	74	47,200	1.57	638
1928–37	Gothenberg, Sweden	birth records	28	27,000	1.04	964
1910–40	Denmark	birth records:				
		all births	193	128,306	1.50	665
		live births	175	121,102	1.45	692
1938–39	Hawaii	birth records	35	18,024	1.98	550
1942	Pennsylvania	birth records	250	202,501	1.23	810
1935–44	Wisconsin	birth records	736	567,509	1.30	770
1942–47	Hawaii	birth records	93	47,153	1.97	507
1943–49	Ontario, Canada	surgical records	695	655,322	1.06	943
1948–50	Pennsylvania	birth records	766	583,690	1.31	762
1948–55	New York (New York City excluded)	birth and death records	1,414	1,242,744	1.14	878
1940–50	Birmingham, England	multiple sources	285	218,693	1.30	767
1951–55	Pennsylvania	birth records	1,269	1,201,976	1.06	947
		all possible sources	1,592	1,201,976	1.32	754
1955	California	birth records	368	313,164	1.18	851
1953–57	Denmark	surgical records	644	393,457	1.64	754
1960–62	Israel	hospital records	26	47,768	.544	1,837

evident that the incidence of these malformations in Israel is lower than average.

For all cleft-types combined, the distribution was approximately even between the sexes. Donahue (*2*) reports that, in his subjects, the incidence of all forms of clefts is higher by 20.4% among boys than it is among girls.

The breakdown for cleft type seems to indicate that in this sample cleft lip alone is more prevalent in girls, while cleft palate alone is more often encountered among boys. These findings differ from those reported by other authors. Loretz and associates (*7*), Beder (*1*), Donahue (*2*), and Hixon (*6*) report that cleft lip alone as well as cleft lip and cleft palate occur more frequently in males than in females, and that cleft palate alone is more prevalent among females. Although the present sample was based on nearly 50,000 live births which occured in hospitals scattered throughout the different parts of Israel, the number of cleft palate births was only twenty-six. Because of the small sample, no statistically valid conclusions could be made. Until such time that a more sizeable sample of clefts has been collected, it is difficult to know whether the trends found in this survey are real or are due to chance.

The age of the mother has frequently been linked with the incidence of clefts in the offspring. Loretz and associates (*7*), Mazaheri (*9*), and McMahon and McKeon (*10*) have found a positive correlation between the incidence of cleft palate and the advance of the mother's age (after age 35). On the other hand, Peer and associates (*11*), Lutz and Moor (*8*), and Grace (*4*) have found no relationship between the age of the mother and the incidence of clefts. Grace (*4*), in a sample of 250 clefts out of a total population of 202,501 cases, found the highest percentage of clefts to be born to mothers falling in the 21–25 year age group. In the present survey, it was found that children with clefts were more often born to mothers in the younger age group (20–24 years) than to older mothers.

In the present study, birth rank did not seem to play a significant role in the appearance of clefts, which conforms with the findings of McMahon and McKeon (*10*).

These findings indicate a comparatively high percentage of additional malformations (27%), as compared with reports by Beder (*1*) (14.5%), McMahon and McKeon (*10*) (15.8%), Loretz and associates (*7*) (18%), and Lutz and Moor (*8*) (25%). These additional malformations affected a) the head and face: microcephalus, encephalopathia, pachimeningitis, facial assymetry, agenesis of the eyes and rudimentary nose, micrognathia, microglossia, and stricture of the oropharynx; b) congenital heart disease; c) extremities: syndactily, polydactily, as well as multiple malformations of the extremities; and d) hydrocele, spina bifida, and others. Stiegler and associates (*12*) cite that the following congenital abnormalities coexist with facial clefts: syndactilysm, polydactilysm,

supernumerary teeth, malformed ears, spina bifida, club foot, and congenital heart disease.

Regarding information about the mother's health condition, it was found that out of the 26 mothers who gave birth to babies with clefts, three suffered from anaemia, two of pregnancy toxaemia, one had rheumatic fever and intestinal ulcers, one was hospitalized in a mental institution, one had two previous artificial abortions which caused disturbances in her menstrual cycle and difficulties in further conceivements, and 13 (50%) had normal pregnancies. No information was available for four mothers. Out of all 26 cases with clefts, only one had a record of a hereditary background. It is quite possible that there were other cases in whom a heritable history could be traced, but that such information was not available from the hospital records. The proportion of one out of twenty-six with a genetic history is lower than the frequency quoted in the literature to be due to genetic factors (20%–30%) (*1, 3, 11*).

The number of live births according to mother's country of origin was estimated by the frequency distributions of these populations in Israel during the years 1960, 1961, and 1962, and was utilized as denominators in calculating the rates of cases with clefts. From this crude estimation, trends are apparent that the incidence of clefts is higher among the populations coming from the Asian countries than for others. Since the population of mothers born in Israel is of greatly mixed geographic descent, it is possible that the relatively high rate of clefts in this group includes mothers whose ancestry originated in the Asian countries. This could not be checked from the birth records, but further studies are being carried out to verify these results.

Summary

To establish the incidence of cleft lip and cleft palate among the Jewish population in Israel, the birth records of six regional hospitals in Israel were examined. The sample comprised 47,768 live births of which 26 showed varying types of cleft lip and cleft palate. The rate per 1,000 live births was 0.544 with a standard error of ± 0.0001, or one in 1,837 cases. No significant differences were found in their distribution between the sexes. Multiple malformations along with cleft lip and/or palate were found in 27% of the cases. Trends were apparent that the incidence of clefts is higher among the groups coming from Asian countries. Further studies are being carried out to verify these results.

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