Clefts in Wisconsin: Incidence and Related Factors

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The literature regarding the incidence of clefts of the lip and/or palate, well summarized by Greene and associates (*6, 7*), characteristically varies in estimates of incidence and in citing possible etiologic factors. Incidence figures vary from one in 276 to one in 2,400 live births (*5, 6, 11*). Factors of etiologic significance have included genetic variables, drugs, radiation, dietary deficiencies, sex, mechanical obstructions in utero, maternal illness during early pregnancy, race, and an assortment of geographic and parental variables possibly affecting the occurrence of clefts.

The present study attempts to explore incidence, etiology, and concomitant problems of children with lip and/or palatal clefts born in Wisconsin from 1943 to 1962. The areas investigated are analyzed under the headings of incidence, birth data, parental variables, selected social variables, and selected geographic variables.

Method

In Wisconsin, reporting of congenital anomalies is required within 24 hours of birth. The birth certificates also provide information regarding sex, length and weight of the child, factors related to pregnancy, labor and delivery, and parental ages, residence, and paternal occupation. The data utilized in this study were obtained primarily from photostatic copies of these birth certificates, which were forwarded by the Wisconsin Board of Health to the Bureau for Handicapped Children, a section of the Wisconsin State Department of Public Instruction. Supplemental information from case files was also obtained from the Bureau for Handicapped Children. As suggested by Greene and associates (*7*), although information obtained from birth records is neither complete nor accurate in detail, the full utilization of such records within their limitations has not been realized.

Although 2,166 cases of clefts occurring over the 20-year period were studied, some categories of information were not universally obtainable, hence the varying totals for the tables presented.

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TABLE 1. Incidence in Wisconsin of lip and/or palatal clefts, 1943–1962. No year of birth was given for one cleft lip, one cleft palate, and two lip and palate clefts.

Year	*Cleft lip*	*Cleft palate*	*Cleft lip and palate*	*Total clefts*	*Ratio of cleft to total live births*
1943	9	23	30	62	1:1041
1944	16	22	29	67	1:913
1945	21	28	34	83	1:733
1946	30	27	44	101	1:734
1947	34	36	59	129	1:646
1948	31	34	52	117	1:687
1949	30	31	55	116	1:710
1950	41	31	39	111	1:740
1951	37	29	54	120	1:730
1952	31	33	38	102	1:872
1953	24	40	42	106	1:831
1954	27	51	39	117	1:780
1955	30	26	48	104	1:884
1956	25	31	76	132	1:708
1957	15	33	39	87	1:1109
1958	21	32	52	105	1:914
1959	33	36	47	116	1:849
1960	46	34	61	141	1:705
1961	29	39	47	115	1:855
1962	41	24	54	119	1:791
				12 facial clefts	
Total	571	640	939	2162	
Mean	28.6	32.5	47.5	108.3	1:812.4
Range	9–46	22–51	29–76	62–141	1:646–1:1109
SD	8.86	3.20	8.50	14.56	111.62

Results and Discussion

INCIDENCE. The total incidence by type of cleft over the 20-year period from 1943 to 1962, as well as for each year within that period, is given in Table 1. The average incidence of clefts is one in 812 live births. Phair (*10*) found the Wisconsin incidence for the decade preceding this study (1933–1942) to be one in 770 live births. The present study indicates a decrease in incidence in the amount of 9.35% in comparison to Phair's findings. The variation in incidence of clefts over the 20 years of this study was not always proportional to the variation in total live births.

With the exceptions of 1950 and 1954, the number of combined lip and palate clefts exceeded isolated clefts of the lip or palate. Over the 20 years studied, 26% of the total were clefts of the lip only, 30% were clefts of the palate only, and 44% were combined clefts.

The incidence of type of cleft by sex is shown in Table 2. Of the 2,157 cases for whom sex was listed, 60% were males, and 40% females. Of the isolated lip clefts, 64.5% were males and 35.5% were females. Isolated palatal clefts occurred somewhat less frequently among males than fe-

TABLE 2. Incidence in Wisconsin of type of cleft by sex, 1943–1962.

Sex	*Cleft lip*		*Cleft palate*		*Cleft lip and palate*		*Total clefts*	
	Number	%	*Number*	%	*Number*	%	*Number*	%
Male	369	64.5	296	46.2	628	66.7	1293	60
Female	203	35.5	345	53.8	313	33.3	861	40
Total	572	26.34	641	29.92	941	43.74	2154	

TABLE 3. Incidence in Wisconsin of type of cleft by month of birth, 1943–1962.

Month	*Cleft lip*	*Cleft palate*	*Cleft lip and palate*	*Total number of clefts*
January	47	51	74	172
February	53	53	79	185
March	53	36	95	184
April	47	51	70	168
May	49	39	79	167
June	40	70	89	199
July	59	56	92	207
August	45	52	65	162
September	46	44	72	162
October	41	53	70	164
November	44	53	75	172
December	44	65	71	180
Total	568	623	931	2122

males: 46.2% to 53.8%. Combined clefts of the lip and palate occurred 66.7% in males and 33.3% in females.

Trends relating to the month of birth were also studied. Table 3 indicates that monthly occurrences were similar, with the possible exceptions of June and July, which tended to be somewhat higher. The hypothesis that no difference exists in the incidence of cleft births among the 12 months for the 20 years studied was tested by means of chi square. The resulting chi square of 13.33 with 11° of freedom was not sufficient to reject the null hypothesis, indicating that month of birth (and hence of conception) did not affect the occurrence of clefts. No trends relative to Wisconsin seasons are apparent, with the possible exception of the higher June–July figure.

Birth Data. Birth data evaluated in this study included the term of pregnancy, the week at which pre-natal medical care began, the length and weight at birth, parity, pre-natal complications of this and previous pregnancies, and complications of labor and delivery. These data are summarized in Tables 4 through 7.

TABLE 4. Cleft births in Wisconsin, 1943–1962: length of gestation, initiation of pre-natal care, length of infant at birth, weight of infant at birth.

Item studied	*Criterion*	*Number reporting*	*At criterion*		*Exceeding criterion*		*Below criterion*	
			Number	*%*	*Number*	*%*	*Number*	*%*
Length of gestation	37–40 weeks	1645	1456	88.5	90	5.5	99	6.0
Initiation of pre-natal care	12th week	1593	431	27.0	496	31.0	666	42.0
Length of infant	18–21 inches	1276	1031	80.8	130	10.2	115	9.0
Weight of infant	5′8″–10′2″	1424	1286	90.3	20	1.4	118	8.3

Term of Pregnancy. Full-term pregnancies, those lasting 37 to 40 weeks, accounted for 88.5% of the cleft births. Births occurring prior to 37-weeks gestation constituted 6%, and those beyond 40 weeks, 5.5% of the cleft births (Table 4).

Prenatal Care. Sixty-nine per cent of the mothers involved in this study were listed as having obtained pre-natal care during the first trimester, more than half of these mothers having been seen by their physicians before the twelfth week of pregnancy (Table 4).

Length at Birth. Approximately 81% of the cleft babies studied fell into the 18- to 21-inch criterion for normal length at birth, with 10% exceeding and 9% falling below criterion (Table 4).

Birthweight. Greene's four state study (7) suggests that babies with clefts are smaller than expected for length of gestation, 14% of his sample being considered premature. Based on a birthweight of less than five and one-half pounds as signifying prematurity, 8.3% of the babies evaluated in the present study would be considered premature. Utilizing the Wisconsin cleft and noncleft birth data from 1958 through 1962, the hypothesis of independence of birthweight (more or less than five pounds eight ounces) and the presence or absence of a cleft was tested (Table 5). The resulting chi square of 2.75 with one degree of freedom is insufficient to reject the null hypothesis, indicating that these data do not demonstrate a difference in birthweights between cleft and noncleft infants.

TABLE 5. Birthweights of Wisconsin infants.

Birthweight	*Cleft births, 1943–1962*		*Cleft births, 1958–1962*		*Noncleft births, 1958–1962*	
	Number	*%*	*Number*	*%*	*Number*	*%*
Under 5 lb. 8 ozs.	118	8.3	43	8.2	30,989	6.2
5 lb. 8 ozs. & over	1306	91.7	484	91.8	453,868	93.8
Total	1424		527		484,857	

TABLE 6. Parity of Wisconsin infants.

Birth order	*Cleft births, 1943–62*		*Cleft births, 1958–62*		*Noncleft births, 1958–62*		*Ratio of clefts to total live births, 1958–62*
	Number	%	*Number*	%	*Number*	%	
1	429	25.9	104	19.88	112,262	23.10	1:1079
2	413	25.0	113	21.60	110,475	22.72	1:978
3	319	19.3	102	19.50	94,362	19.41	1:925
4	196	11.8	79	15.10	67,842	13.95	1:869
5	101	6.1	43	8.22	42,163	8.64	1:981
6	75	4.5	34	6.50	25,090	5.16	1:738
7	41	2.5	12	2.29	15,014	3.08	1:1251
8+	81	4.9	36	6.88	18,847	3.87	1:524
Total	1655		523		486,055		

Parity. Order of pregnancy was explored, revealing that the cleft infants studied occurred anywhere from the first through the fifteenth pregnancy, the average being slightly over the third. Table 6 presents the numbers and percentages of cleft and normal births, as well as the incidence of clefts by birth order. The hypothesis of the independence of birth order and the presence or absence of cleft was tested. The resulting chi square of 18.28 with seven degrees of freedom is sufficient to reject the null hypothesis. These data indicate, then, that there is a relationship between birth order and the occurrence of clefts, with a considerable increase in the incidence of clefts beyond the seventh pregnancy (the ratio of observed to expected occurrence was 1.77).

Complications of Pregnancy, Labor, and Delivery. Of the 2,166 certificates studied, 783 reported complications of pregnancy, labor, and delivery (as noted and reported by the attending physician). Complications of labor include abnormalities of membrane rupture, placenta previa, prolonged or excessively hard labor, functional uterine problems (such as inertia or pelvic arrest), abnormal fetal position, excessive fetal size, prophylaxis (the necessity of taking action to avoid difficulty), and complications not otherwise identified by name. Delivery complications include breech, forceps, Caesarean, difficult, and operative (type not specified) deliveries. Complications of pregnancy include such difficulties as toxemia, hemorrhage, maternal disease and accidents, and complications otherwise not identified by name. Table 7 presents a breakdown of the numbers and types of complications by the type of cleft.

The greatest number of women experiencing difficulties were mothers of children having combined clefts of the lip and palate, which is in keeping with the relative size of the combined clefts group. Forceps and Caesarean deliveries accounted for 92.5% of the complications of delivery. Of special interest is the fact that 106 mothers for whom pre-

TABLE 7. Complications of pregnancy, labor, and delivery occurring in Wisconsin infants born with lip and/or palatal clefts, 1943–1962. Previous abortions were reported only on birth certificates from 1959 through 1962.

Category studied	*Cleft lip*		*Cleft palate*		*Cleft lip and palate*		*Number reporting in each category*
	Number	%	*Number*	%	*Number*	%	
Labor complications	29	26.4	31	28.2	50	45.4	110
Pregnancy complications	12	26.7	15	33.3	18	40.0	45
Delivery complications	103	28.3	91	25.0	170	46.7	364
Previous abortions	33	31.3	31	29.3	42	39.6	106
Previous stillbirths	10	20.8	11	22.9	27	56.3	48
Previous live births dying later	24	21.8	35	31.8	51	46.4	110
Total	211		214		358		783

vious abortions (pregnancies terminating prior to 20-weeks gestation) were reported had experienced a total of 168 abortions. This number of abortions, recorded in only four years, suggests the need for further study of abortion as a factor in the occurrence of clefts, especially as it may relate to the occurrence of lethal hereditary factors (*8*) or to maternal serum protein disturbances (*9*).

Chi square tests were utilized to test the hypotheses of independence of cleft type and the various complications of pregnancy, labor, and delivery studied. None of the chi square values was sufficiently great to reject the null hypotheses, indicating that for these data the type of cleft was not related to the complications explored. The pre-natal and perinatal histories of the mothers of the infants included in this study do not, therefore, suggest any reasons for differences in the incidence of the different cleft types.

Multiple Births. Multiple births were investigated, revealing 40 twins but no other multiple births among the cleft births recorded during the 20 years studied. Of the 40 twin births, the cleft infants were equally distributed among the first- and second-born, suggesting that birth order in a multiple birth is unrelated to the occurrence of clefts. The certificates did not establish which twins were monozygotic or dizygotic. Since clefts occurred in both twins in only four cases, none being identical clefts, none of the twins were concordant for cleft type. Twin births accounted for only 1.8% of the 20-year total of clefts.

Multiple Anomalies. Table 8 summarizes the occurrence of additional anomalies in the babies studied. *Other anomalies,* a group of abnormalities occurring only once or twice and not indicated in the literature as coincidental with clefts, constituted the largest category. Consistent with the earlier literature, the most frequent anomaly was club foot (11.5%). Club foot, along with congenital heart defects (9.9%), atresia of

TABLE 8. Additional anomalies occurring in Wisconsin infants born with lip and/or palatal clefts, 1943–1962. Included in the total are 25 subjects who had two or more additional anomalies, totaling 58 additional anomalies in all.

Anomaly	*Cleft lip*	*Cleft palate*	*Cleft lip and palate*	*Facial cleft*	*Total*
Club foot/feet	1	11	8		20
Spinal anomaly or Spina bifida		2	1		3
Supernumerary fingers	1	3	3		7
Supernumerary toes	1		4		5
Webbed fingers		4	5		9
Webbed toes		3			3
Absence of leg			2		2
Absence of foot			1	1	2
Absence of arm	1	1	2	1	5
Absence of hand or fingers	2		2	1	5
Atresia or other ear deformity	2	7	4		13
Pierre Robin syndrome	1	13			14
Eye defects	2	4	3		9
Heart defects	2	6	9		17
Mongolism	1	4	1		6
Other anomalies	6	28	16	3	53
Total	20	86	61	6	173

the ear (7.5%), and Pierre Robin syndrome (micrognathia) (8.1%) constituted somewhat more than one-third of the total additional anomalies.

Infants having palatal clefts accounted for 49.7% of the group with multiple congenital anomalies, whereas 35.5% occurred in those having combined clefts of the lip and palate. Infants with clefts of the lip constituted the remaining 14.8% of those having additional anomalies. The occurrence of additional anomalies is obviously not proportional to the size of the three cleft groups. The hypothesis that the occurrence of additional defects is independent of the type of cleft was tested (Table 9). The resulting chi square of 45.36 with two degrees of freedom is sufficient to reject the null hypothesis, and to conclude that a real relationship exists between the type of cleft the infant has and the occurrence of addi-

TABLE 9. Presence or absence of additional congenital anomalies among Wisconsin infants born with lip and/or palatal clefts, 1943–1962.

Additional anomalies	*Cleft lip*		*Cleft palate*		*Cleft lip and palate*		*Combined groups*	
	Number	*%*	*Number*	*%*	*Number*	*%*	*Number*	*%*
Present	20	3.49	86	13.42	61	6.48	167	7.75
Absent	552	96.51	555	86.58	880	93.52	1987	92.25
Total	572		641		941		2154	

TABLE 10. Ages of Wisconsin mothers at birth of child.

Maternal age	*Cleft births, 1943–62*		*Cleft births, 1958–62*		*Noncleft births 1958–62*		*Ratio of cleft to total live births, 1958–62*
	Number	*%*	*Number*	*%*	*Number*	*%*	
19 and under	128	7.4	52	9.89	35,880	9.19	1:691
20–24	593	34.1	180	34.22	130,718	33.48	1:727
25–29	446	25.6	135	25.67	106,321	27.23	1:788
30–34	334	19.2	102	19.39	68,627	17.58	1:674
35–39	175	10.1	41	7.79	37,420	9.58	1:914
40 and over	63	3.6	16	3.04	11,387	2.91	1:713
Total	1739		526		390,353		

tional congenital anomalies. The group having clefts of the palate only had more anomalies (the ratio of observed to expected occurrence was 1.73), and the cleft lip only and combined clefts group had fewer anomalies (ratios were .45 and .77, respectively) than would be expected under the hypothesis of independence.

PARENTAL VARIABLES. The parental variables assessed in this study include maternal and paternal ages at the time of the birth and the presence of clefts and other anomalies in the family.

The mean maternal and paternal ages were 27 and 30 years, respectively. The maternal age range was 14 to 50 years and the paternal age range was 17 to 62 years. In only a very few cases were the mothers older than the fathers. Maternal and paternal ages, charted in five-year groupings, are presented for the parents of cleft infants in Tables 10 and 11. Also presented in these tables are parental age distributions for mothers (1958–62) and fathers (1959–62) of cleft and noncleft infants, and the incidence of clefts by maternal and paternal age.

Maternal Age. The largest maternal age group, 20 to 24 years, pro-

TABLE 11. Ages of Wisconsin fathers at birth of child.

Paternal age	*Cleft births, 1943–62*		*Cleft births, 1959–62*		*Noncleft births 1959–62*		*Ratio of cleft to total live births, 1959–62*
	Number	*%*	*Number*	*%*	*Number*	*%*	
19 and under	23	1.34	10	2.27	7,242	1.90	1:725
20–24	339	19.70	96	21.77	84,019	22.11	1:876
25–29	520	30.21	139	31.52	114,359	30.10	1:824
30–34	393	22.84	94	21.31	87,631	23.06	1:933
35–39	231	13.42	56	12.70	51,928	13.66	1:928
40 and over	215	12.49	46	10.43	34,702	9.13	1:755
Total	1721		441		379,881		

duced 34% of the cleft infants. A total of 60% of the cleft infants were born to mothers in the 20- to 29-year decade. Women 35 years or older produced 13.7% of the cleft infants. The hypothesis of independence of maternal age and the presence or absence of a cleft in the infant was tested using the cleft and noncleft births for 1958 through 1962. The resulting chi square of 3.59 with five degrees of freedom was insufficient to reject the null hypothesis. These data do not support the theory that clefts may occur more frequently in children of mothers at either extreme of the age continuum.

Paternal Age. The largest paternal age group, 25 to 29 years, produced 30% of the cleft infants. A total of 50% of the cleft infants were born to the fathers in the 20- to 29-year decade. Men 35 years or older produced 26% of the cleft infants, or about twice as many as women in the same older age group. The hypothesis of independence of paternal age and the presence or absence of a cleft in the infant was tested using the cleft and noncleft births for 1959 through 1962. The resulting chi square of 2.32 with five degrees of freedom was insufficient to reject the null hypothesis. The data do not support the theory that clefts may occur more frequently in children of fathers at either extreme of the age continuum.

Familial Anomalies. The presence of familial anomalies, although not recorded on birth certificates, is obtained where possible by the Bureau for Handicapped Children. Inspection of the distribution of familial anomalies (Table 12) reveals that clefts occurred more than 12 times as frequently as other anomalies (149 to 12). Familial clefts were listed in 6.9% of the total cases studied. Siblings accounted for 73% of the familial clefts. Using the 6.9% familial clefts occurrence, one cleft in every 14.5 births would be expected. The previous literature suggests a higher familial incidence than the figure obtained in the present study, which must be considered a minimal estimate. Had a standard pattern been used in questioning the parents and other relatives regarding familial

TABLE 12. Incidence of clefts and other anomalies in families of Wisconsin infants born with lip and/or palatal clefts, 1943–1962.

Relationship to infant	*Number reporting*	*Type of cleft in infant*		
		Cleft lip	*Cleft palate*	*Cleft lip and palate*
Maternal relatives cleft	20	2	7	11
Paternal relatives cleft	16	4	2	10
Unstated relatives	4	2		2
Sisters	31	7	6	18
Brothers	63	18	9	36
More than one sibling	15	1	8	6
Total familial clefts	149	34	32	83
Noncleft anomalies	12	4	4	4

TABLE 13. Numbers of reported clefts in relatives of Wisconsin infants born with lip and/or palatal clefts, 1943–1962.

Clefts in relatives	*Type of cleft in infant*							
	Lip only		*Palate only*		*Lip and palate*		*Combined groups*	
	Number	*%*	*Number*	*%*	*Number*	*%*	*Number*	*%*
Number reporting clefts.....	34	5.94	32	4.99	83	8.82	149	6.92
Number not reporting clefts..	538	94.06	609	95.01	858	91.18	2005	93.08
Total	572		641		941		2154	

clefts, a greater familial incidence may have been ascertained. Nonetheless, the obtained figures support the concept that heredity is of etiologic significance in clefts of the lip and/or palate.

The hypothesis of independence of type of cleft in the infant and occurrence of cleft in the family was tested (Table 13). The resulting chi square of 9.83 with two degrees of freedom is sufficient to reject the null hypothesis. Infants having clefts of the lip or clefts of the palate had fewer relatives with clefts than expected under the hypothesis of independence (ratios of occurrence were .86 and .72, respectively). Infants having combined clefts of the lip and palate had more cleft relatives than expected (the ratio of occurrence was 1.28). These findings, along with the differences in sex ratios and in the occurrence of additional anomalies, tend to support the hypothesis of etiological distinction among the various types of clefts (*2, 3, 4, 8*).

ETHNIC DIFFERENCES. Table 14 presents the incidence of clefts by race. As would be anticipated from the great predominance of Caucasians in the state population, the majority of clefts (98.9% for whom race was listed) occurred among Caucasian infants. Six clefts occurred among Negroes, resulting in an incidence of one in every 5,607 Negro births,

TABLE 14. Incidence in Wisconsin of cleft births by ethnic group, 1943–1962.

Ethnic group	*Reporting clefts*		*Ratio of cleft to total live births*
	Number	*%*	
Caucasian.....	1740	98.90	1:960
Indian.....	8	.45	1:1265
Mixed.....	5	.30	1:296
Negro.....	6	.35	1:5607
Oriental.....	1	—	—
Total.....	1760		

TABLE 15. Rural and urban distribution of Wisconsin births, 1943–1962. Urban denotes counties in which the majority of the population resides in communities of 2,500 or more people. Rural denotes a majority residing in communities of less than 2,500.

Maternal residence	*Cleft births*	*Percentage of cleft births*	*Noncleft births*	*Percentage of noncleft births*
Rural (59 counties)	896	41.3	616,890	35.5
Urban (13 counties)	1270	58.7	1,117,306	64.5
Total	2166		1,734,196	

and supporting previous reports of decreased incidence of clefts in Negroes. Five cases of clefts occurred in babies of mixed (Caucasian and Oriental or Indian) racial background, resulting in an incidence of one in 296 births, and lending some support to previous findings regarding mixed races. Eight clefts, resulting in an incidence of one in 1,265 births, occurred among American Indians.

SELECTED SOCIAL VARIABLES. Maternal residence and paternal occupation were investigated.

Rural-Urban Residence. Rural versus urban maternal residence was explored to determine its relationship to the occurrence of clefts (Table 15). Although only 13 of the 72 counties in Wisconsin are considered urban (where the majority of the population resides in communities of 2,500 or more people) these 13 counties account for the majority of the state population and of the total cleft and noncleft births. Interestingly, the proportions of cleft and noncleft births in rural and urban areas are not equal. The hypothesis of independence of maternal rural-urban residence and the presence or absence of a cleft in the infant was tested. The resulting chi square of 31.69 with one degree of freedom is sufficient to reject the null hypothesis. Rural counties account for a greater proportion of clefts than would be expected under the hypothesis of independence (the ratio of occurrence was 1.16). Although real, the relationship between rural residence and the occurrence of cleft is weak enough so that one would scarcely predict cleft offspring on the basis of maternal residence. This relationship, however, may reflect genetic factors that are influenced by the possibility of greater consanguinity or greater social and economic security for individuals with clefts, and hence a greater cleft population, in rural areas.

Paternal Occupation. Fathers' occupations were classified according to 13 categories selected from the United States census reports and the *Dictionary of Occupational Titles* (Table 16). A wide range of socioeconomic levels is probably incorporated in each category, since the level of each individual's occupation is ambiguously recorded on birth certificates. Agricultural, building and construction, and production

TABLE 16. Occupations of fathers of Wisconsin infants born with lip and/or palatal clefts, 1943–1962.

Paternal occupation	*Number reporting*	*Percentage*
Agriculture	335	18.5
Building and construction	284	16.0
Production workers	256	14.5
Skilled mechanics or technicians	175	10.0
Service industry	180	10.5
Middle management	58	3.2
Management	41	2.2
Transportation	124	7.0
Professional	110	6.1
Self-employed	75	4.2
Student	60	3.3
Military	57	3.2
Office work	24	1.3
Total	1779	

workers account for 49% of the total paternal occupations listed. Professional and managerial positions constitute only 8.3% of the total.

SELECTED GEOGRAPHIC VARIABLES. The distribution of cleft births among the counties within the state was explored, along with the incidence within each county. As anticipated, more clefts were found in the heavily populated counties, with only few exceptions. Those counties showing high or low incidence of clefts are scattered throughout the state. No pattern other than population size could be established to explain the incidence of clefts.

Glaciation. Glaciation (decreased iodine) and nonglaciation throughout the state, considered as possibly relating to the presence of iodine in the maternal diet, and consequently to the occurrence of clefts (*1*), was examined. Table 17 indicates that the highest incidence of clefts occurred in the nine partly glaciated counties. The lowest incidence occurred in the 50 glaciated counties. These findings disagree with those of Brown (*1*), who found a higher incidence in glaciated than in nonglaci-

TABLE 17. Incidence of cleft and noncleft births in Wisconsin according to glaciation of the counties of maternal residence, 1943–1962. (Glaciated counties were defined as those which were invaded by the continental glacier during the ice age.)

Glaciation	*Number of cleft births*	*Number of live births*	*Ratio of clefts to total live births*
Nonglaciated (13 counties)	214	153,316	1:716
Partly glaciated (9 counties)	390	269,654	1:691
Glaciated (50 counties)	1,562	1,313,392	1:841

ated areas. The present findings suggest that the relationship between glaciation (decreased iodine) and the occurrence of clefts is questionable.

Summary

This study analyzed birth certificate data and supplemental information obtained from the files of the Bureau for Handicapped Children, State of Wisconsin. A total of 2,166 cleft lip and/or palate births occurring from 1943 to 1962 were investigated with regard to incidence, birth data, and selected parental, social, and geographic variables.

Specifically it was found that: a) The total incidence of clefts during the 20-year period was one in 812 live births. b) Of the total group, 26% were lip-only clefts, 30% palate-only clefts, and 44% combined clefts of the lip and palate. c) Males accounted for 60% and females 40% of the births. d) Males outnumbered females by approximately a 2:1 ratio in clefts of the lip only and in combined clefts of the lip and palate, but the ratio of males to females was 46:54 for clefts of the palate only. e) The occurrence of clefts is independent of month of birth (and hence conception), birthweight of the infant, maternal age, paternal age, and glaciation of the county of maternal residence. f) The occurrence of clefts seems related to birth order, with considerably more cleft palate births than expected occurring at the eighth pregnancy and beyond; to rural-urban maternal residence, with considerably more clefts than expected occurring in rural mothers; and to race, with incidence of clefts being higher in Caucasians than in Negroes or Indians. The highest incidence occurred in mixed races.

g) The type of cleft (lip only, palate only, combined lip and palate) is independent of complications of labor, delivery, or of this and previous pregnancies. h) The type of cleft appears to be related to the presence or absence of additional congenital anomalies in the infant, with cleft palate-only infants having considerably more anomalies than expected and cleft lip-only and combined cleft infants having fewer than expected, and to the occurrence of clefts in the family, with infants who have combined clefts having more cleft relatives than expected and cleft lip-only or palate-only infants having fewer than expected. i) The occurrence of clefts in the family of cleft infants was considerably greater than would be expected from general population incidence figures, which supports the significance of heredity as an important etiologic factor. j) The relatively high incidence of previous spontaneous abortions in mothers of these cleft infants suggests the need for further investigations, especially as it may relate to lethal hereditary factors or maternal hormonal-metabolic or serum protein disturbances.

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