Cleft Palate in Montana: A 10-Year Report

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The purpose of this paper is to provide a review of the data collected on 363 babies with clefts born live in Montana over a ten-year period from January 1955 to January 1965.

The major source of information for identifying subjects was birth certificates. Congenital anomalies are listed on the birth certificates in Montana, and a copy of a certificate indicating a cleft is sent to the Cleft Palate Program. Although all clefts and other pertinent information are not always listed on the certificate, most children with clefts born in Montana receive care under this Program and case records are a source of additional information. The Division of Vital Statistics has furnished other pertinent material used in this paper. Of the 363 live births with clefts born in this 10-year period, 24 died quite soon after birth, 6 moved out of the state immediately, thus leaving 333 known in the state. Of these, 289 or 86.7% were registered in the Program.

Medical Information

DEATHS. No deaths have occurred as a result of a surgical procedure performed by one of the Program surgeons. Although one 13-day-old baby died during lip surgery performed by a physician who was not a Program surgeon, autopsy revealed an enlarged thymus and it is possible that congenital anomalies contributed to this death.

Of the total 363 babies, 42 or 11.5% have died. Loretz' figures show 12% of the babies in his California study died before 6 months of age (*9*). Fourteen deaths in our study occurred within one week of birth, 6 between one week and one month, 15 between one month and six months, and 7 from six weeks to four years of age.

Thirty-seven or 88.09% of the 42 babies who died had identifiable congenital anomalies which probably contributed to death. Prematurity and hyaline membrane disease were stated as the cause of death in two cases. Inconclusive information was available on one case and two children died of illnesses unconnected with their clefts.

Our findings support the contention that cleft palate is a more serious defect than cleft lip and lip and palate. For example, only one-third of the total sample had cleft palate only, yet almost one-half (45%) of those babies who died had cleft palate.

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The inadequacy of birth certificates only for such studies was apparent in studying the deaths of these babies. In ten of the 37 cases where congenital anomaly probably contributed to the death of the child, the anomaly was not listed on the birth certificate. If one were listing anomalies from birth certificates only, almost 25% of the anomalies severe enough to contribute to death within one month was omitted. (We became aware of the anomalies from information on death certificates and from direct contacts between the families and other professional persons.)

Birth Weights. Information is available about the birth weights of 312 of these 363 babies. Of the total group, 46 or 14.7% had birth weights of 5 pounds 8 ounces or less. There was no statistical difference between the birth weights of babies with cleft palate only and those with cleft lip and palate and cleft lip. Over this same period, 7.5% of babies born live in Montana without clefts had birth weights of 5 pounds 8 ounces or less. This high incidence of prematurity (defined by birth weight) has been noted by others (*3, 5, 6*).

Twinning. Reports on twin births with clefts are in the literature (*4, 5*). In this ten-year period, eight sets of twins were born, or 2.2% of the 363 were twin births. In Montana over this same period, 1.8% of all births were twin. In all eight cases in the cleft group, only one twin had a cleft and none of the sets was identical. In four of the cases, the lip only was involved, two had palate clefts only, and two were of both lip and palate.

Cleft Type and Sex. Information from our studies about cleft type is in essential agreement with those facts reported by a number of other investigators (*4, 5, 6, 9, 11*). For example, males predominate in the whole group—198 or 54.6%. Of the total group, 121 or 33.3% of the clefts are palatal clefts, 88 or 24.2% are lip clefts, and 154 or 42.4% are cleft lip and palate. Regarding sex distribution, 70 or 57.8% of the palate only clefts, 34 or 38.6% of the lip clefts, and 61 or 39.6% of the lip and cleft lip and palate group were female. Identifying information about side of cleft, in the lip and palate group, was not available for 19 or 5.2% of the babies. However, of the identified groups, bilateral clefts made up 50 or 32.4% of the total lip cleft group and, of the unilateral clefts, 53 or 62.4% were on the left side.

Other Facial Anomalies. Other facial anomalies were reported during this 10-year period: one case of lip pits, four cases of rudimentary or "healed" cleft lips, one cleft tongue, one lateral cleft (macrostomia), two mandibular alveolar clefts (1 with the cleft tongue, also), seven submucous palatal clefts of a degree severe enough to require care, five Pierre-Robin syndromes (in addition, a tendency toward this was reported in other cases), one Treacher-Collins syndrome, and one central cleft. Peer and others (*14*) noted that babies with central clefts did not live long and this was true in this case.

TABLE 1. Distributions of cleft type and incidence of clefts reported in the family. (Entries are percentages and numbers are in parentheses.)

	cleft palate only N = 121	*cleft lip or cleft lip and palate N = 242*
per cent of clefts in immediate family or history	20.57% (25)	26.03% (63)
per cent of clefts which were immediate family clefts (parents-siblings)	32.00% (8)	44.40% (28)
per cent of immediate family clefts in mother	50.00% (4) (all cleft palate)	7.14% (2) (cleft lip and palate or lip)
per cent of immediate family clefts in father	0.00% (0)	28.57% (8) (all cleft lip and palate)
per cent of immediate family clefts in siblings	50.00% (4) (2 cleft palate)	64.28% (18) (16 or 88.8% lip or lip and palate)

CLEFTS IN THE FAMILY HISTORY. Clefts were reported in the family history of 88 or 24.2% of the total group. A total of 63 or 26.03% of the babies in cleft lip and cleft lip and palate groups combined and 25 or 20.6% of the babies with cleft palate only had clefts reported in their history. No more detailed information about the history of clefts beyond the immediate family (parent, siblings) is available. It seems appropriate to point out, however, that pedigrees for Montana families can rarely be traced further than the child's great grandparents. (Montana has been a territory for only 100 years and a state for a little over 75; most family ties with relatives in the East are weak.)

The immediate family history revealed an interesting pattern of distribution and patterning as shown in Table 1. Distribution of the clefts in the history of children with cleft palate only showed that 32% of these occurred in the immediate family of the child with 50% of those in the mother, 50% in the siblings, and none in the father. All of the mothers who had clefts had cleft palate only and one-half of the siblings who had clefts also had cleft palate only. Of the clefts reported in the families of the 63 babies with cleft lip and lip and palate, 28 or 44.4% were in the immediate family. The distribution differs from cleft palate only, however. Eighteen or 64.28% of the family clefts were in a sibling, 8 or 28.57% of the father and 2 or 7.14% in the mother. All the fathers' clefts are of the lip or lip and palate, and 92.3% of the clefts of siblings were cleft lip or cleft lip and palate. Spriestersbach also noted this high inheritance of similar types of clefts (*15*).

OTHER CONGENITAL ANOMALIES IN CHILD AND FROM HISTORY. Our studies show that 132 or 36.3% of the 363 babies in this study have congenital anomalies reported in the child or from the family history or both. We should point out that our studies include the babies with anomalies who have died and that information of this type may not be

included in studies of some researchers. This may account in part for the considerable disparity among investigators concerning reports on anomalies in cleft cases (*4, 5, 6, 10*). Of the babies with cleft palate only, 45 or 37.1% have anomalies reported in the child, 20 or 16.5% in the family history, and 7 or 5.7% in both child and family history. In the case of cleft lip and cleft lip and palate, anomalies were reported in 49 or 20.3% of the children, in 31 or 12.8% of the families, and in 6 or 2.4% of both child and family history. In most cases an anomaly in the member of a family extends only to great grandparents, as previously indicated.

Social Information

ADOPTIONS. Of the 363 babies born during this period, 11 are adopted or in foster home care. Our experience seems to show that a cleft is not necessarily a deterrent to adoption since 8 have been adopted and 3 are in foster homes. The 3 children in foster homes had not been released for adoption until they were past babyhood; perhaps serious efforts to seek adoptive parents may not have been made in these cases. Six of the adopted children had cleft lip and palates and 1 of them also had phocomelia. Two children were of Indian descent and adopted by white parents.

INSTITUTIONALIZED AND SEVERELY RETARDED CHILDREN. Ten children of this group were institutionalized because of severe retardation. At the present time, there appear to be 11 retarded children who are at home, but who might well be institutionalized before they reach adulthood. A total of 21 children out of the 363, then, appear severely retarded. Of these children, 1 has a central cleft, 11 have cleft palate, and 7 have cleft lip or cleft lip and palate. These studies again emphasize the seriousness of cleft palate only as far as other accompanying problems obtain.

Factors Affecting Montana's Incidence of Cleft Palate

As previously reported (*16*), Montana's incidence of cleft palate is high when compared with reports of other studies. As shown in Table 2, there has been one cleft palate birth in every 465 live births in

TABLE 2. Incidence of live births with clefts in Montana listed by race from 1/1/55 to 1/1/65 (by occurrence).

race	*birth rate in Montana*	*number with clefts*	*incidence ratio*
Caucasian	156,847	325	1:483
Indian	10,724	38	1:282
Negro	580	0	0
Chinese	65	0	0
Japanese	261	0	0
other	298	0	0
total	168,865	363	1:465

Montana over this 10-year period. This can be compared with reports from other states (*5*, *7*, *13*), of an incidence rate of roughly one cleft birth in 700 births.

General Factors. Several factors appear to be operating in this incidence figure. One is the high Indian incidence of clefts also reported by us previously (*16*) and, in this study (Table 2), one in 282 births. However, even when Indian populations are eliminated, the incidence rate is still one cleft in 486 live births in the state. Obviously, other factors are also affecting the incidence figure.

One factor that may well affect Montana's incidence even more than the high incidence of clefts in the Indian population is the extremely small Negro population in the state (Table 2). A number of researchers have reported that the occurrence of clefts in the Negro population is considerably less than in the Caucasian race (*8*, *1*) and so general incidence rates for populations which include large groups of Negroes would tend to be depressed. In Montana, only 580 or .003% of the births were Negro during the ten-year period, with other races making up an even smaller proportion of the total. This population make-up, undoubtedly, contributes to the high incidence rate reported in the state.

In addition, the high incidence figure in Montana may reflect the relatively high degree of completeness in reporting clefts, since incidence figures are obtained from a combination of case records and birth certificates for our studies. Birth records do not include all clefts. As Milham (*12*) pointed out, 18.2% of the children in a study from three upstate New York Hospitals would have been missed if vital records were used. Further, he reports that in the New York Department of Health only 70% of the birth defects were on the certificate (*12*). Ivy points out a 16% error in under-reporting in reviewing Pennsylvania's vital statistics (*7*). In 1963 (*16*), we reported that 5% of the children born during a 6-year period in Montana did not have their clefts listed on the birth certificates. Our studies now show 17.3% (63) clefts not on birth certificates. In this study, had we reported incidence of clefts from birth certificates only, the incidence rate would have been 1 in 593 live births in the state over the 10-year period instead of 1 in 465. The incidence figures we have published in the past have not been as high as the present figure and the difference probably represents, for the most part, much of this group whose clefts were not on the certificate and for whom information has now been slowly collected.

Types of Clefts Not Reported on Birth Certificates. Of the 63 clefts not reported on the birth certificates, 36 were clefts of the palate. Thirty-one were rather extensive clefts and 5 were submucous or relatively slight clefts of the soft palate. Twelve were cleft lip only and 5 were rudimentary or "healed" lip clefts. Eight of the clefts were cleft lip and palate, although one of these births was not reported to Vital Statistics. Two were a lateral cleft and a mandibular cleft. Even if we assume that "rudimentary" lip clefts, submucous, relatively slight pala-

TABLE 3. Birth months of live births with clefts compared with birth months of live births without clefts in Montana from 1/56 to 1/65. (The number of cleft births is in parentheses.)

month	*percentage of live births in Montana*	*percentage of cleft births*
January	7.98%	6.3% (21)
February	5.52%	10.6% (35)
March	8.44%	10.6% (35)
April	8.01%	7.29% (24)
May	8.64%	6.3% (21)
June	8.60%	10.03% (33)
July	8.84%	6.07% (20)
August	8.62%	7.9% (26)
September	8.60%	8.51% (28)
October	8.59%	10.3% (34)
November	7.90%	9.4% (31)
December	8.20%	6.3% (21)

tal clefts, and the lateral and mandibular clefts might have reasonably been overlooked at birth, there still remain 50 or 14% of relatively severe clefts which were not indicated on birth certificates.

Influencing Factors

ALTITUDES. The question of whether altitude affects the occurrence of clefts was investigated by assigning an average altitude for each county. In no county was the average at variance with the various towns in it for more than 700 feet; most towns varied in altitude only 200 to 300 feet from the county average. In one county, the altitude of the largest city (a city of 55,000) was used as the county average. Altitudes ranged from 1934 feet to 5890 feet. The distribution of cleft palate births was essentially comparable for all altitudes.

BIRTH MONTH. Several investigators have reported data about the seasonal occurrence of clefts (Table 3). Ingalls was quoted in a popular magazine as stating that the highest number of children with clefts were born in December, January, and March, with the smallest number in June and July (*2*). Our findings agree with him that March is a month of high incidence and that July is a month of low incidence. However, in our findings, December and January are months of low, not high, incidence and June, which he quotes as a low month, is a high one in Montana. Ingalls had reported no statistical significance in seasonal occurrence (*6*). Other investigators do not agree upon this finding and the importance of seasonal occurrence is not yet known (*4, 5, 10*). As shown in Table 3, the seasonal birth rate for all babies in Montana over a nine-year period varied only 1.32 from the highest month (July) to the lowest month (February). The cleft palate births vary 3.99 from

TABLE 4. Comparison of maternal age at birth for live births with and without clefts in Montana from 1/59 to 1/65. (Entires are percentages and numbers are in parentheses.)

groups	*maternal age in years*		
	under 20	*20 to 39*	*40 to 50*
all Montana babies.....	14.22% (14096)	82.23% (82468)	2.53% (2508)
babies with cleft palate only (89).....	12.35% (11)	82.02% (73)	5.61% (5)
babies with cleft lip and palate (134).....	17.91% (24)	78.35% (105)	3.73% (5)
total clefts (223).....	15.69% (35)	79.82% (178)	4.48% (10)

the highest months (February and March) to the lowest month (July). This means that the lowest month for all births (February) is one of the highest months for cleft births. Whether this is the effect of small numbers of cleft births or has significance is not known.

MATERNAL AGE. Data regarding maternal age at birth of child are presented in Table 4 for 223 of the cleft subjects and for all babies born in Montana during a six-year period (1/1959 to 1/1965). The major difference between the two groups is that a larger number of mothers of babies with clefts are 40 years of age and older than are mothers of babies in the general population. A Utah study found that the younger group of mothers had fewer babies with clefts (*17*). This was true of the babies with cleft palate only in our study, but not the babies with cleft lip and palate.

Summary

Of the 363 babies born live with clefts in Montana over a 10-year period, 11.5% died before 4 years of age, in most cases of associated congenital anomalies. Cleft palate only was associated with a higher death rate, a higher rate of mental retardation, and a higher incidence of anomalies than cleft lip and palate. A greater incidence of prematurity is noted in babies with clefts than of all other live births in Montana. Clefts occurred in histories of 24.2% of the whole group, with a larger percent of clefts occurring in the families of babies with cleft lip and lip and palate. Suggested factors for Montana's high rate of clefts (1:465) are the high Indian incidence rate, the extremely small Negro population, and the relative completeness of reporting. A larger percent of mothers 40-year-of-age-and-older than would be expected had babies with clefts, with more of these babies having cleft palate. Other studies are presented concerning the effect of altitude on clefting, seasonal occurrence of births, twinning, occurrence of cleft-associated problems, adoptions, and a study of the type of clefts not reported on birth certificates.

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