Death Certificates as Supplementary Sources of Information in Epidemiologic Studies of Clefts

MURIEL MACKEPRANG and SYLVIA HAY

BIRTH CERTIFICATES, because of their availability at relatively low cost, have been widely used in the study of certain congenital malformations. They serve particularly well in epidemiologic studies of cleft lip and palate because these malformations are easily diagnosed at birth and are among the most common of serious congenital defects. Although clefts are underreported on birth certificates by about 20 percent, there is apparently little, if any, selectivity in reporting (*1*).

Information on age of parents, sex of affected child, date and place of birth, and birth weight are some of the variables of epidemiologic interest that can be assumed to be recorded with reasonable accuracy on birth certificates of children with clefts. Other items, such as the presence of associated anomalies, may not be as well reported on birth certificates, which are, despite their potential value in medical research, principally legal documents, considered by some physicians to be appropriate only for officially recording births.

Death certificates are also primarily legal documents, but they, too, contain many items of interest to epidemiologists and have been frequently used in the study of diseases.

Method and Materials

Matched birth and death certificates of 1,838 infants who were born between 1956 and 1965 with a cleft lip or a cleft palate and who died before 1967 have been recently used to study the causes of death among children born with clefts (*2*). Data collected for that study also provided us with a means of assessing the death certificates as sources of information concerning malformations. Specifically, we wished to discover (*a*) how well clefts were reported on death certificates, (*b*) how much agreement existed between the specific diagnoses (type of cleft) reported on the two kinds of certificates, (*c*) the effect at death on the completeness of reporting clefts on death certificates, and (*d*) what additional information on congenital malformations could be obtained through the study of death certificates.

Punchcards were prepared that contained selected data from each pair of matching birth and death certificates. All congenital malformations recorded on either one or both certificates were coded according to A Classification of Congenital Malformations (*3*).

The clefts were classified by the following diagnoses: isolated cleft lip, combined cleft lip and palate, isolated cleft palate, and Pierre Robin syndrome. Included in these categories were a few cases with ambiguous diagnoses, such as deformity, dysplasia, or defect of the lip or palate, that were assumed to be clefts. In addition to diagnosed cases of Pierre Robin syndrome, cases of cleft palate associated with micrognathia and glossoptosis were redefined as Pierre Robin syndrome and removed from the category of isolated cleft palate.

Results and Discussion

Clefts were reported on 737, or 40 percent, of the 1,838 death certificates (table 1). Three-fourths of the reported clefts were the same type

Miss Mackeprang and Miss Hay are statisticians, Division of Dental Health, Bureau of Health Manpower Education, National Institutes of Health, San Francisco. Tearsheet requests to Miss Sylvia Hay, Dental Health Center, Division of Dental Health, 14th Ave. and Lake St., San Francisco, Calif. 94118.

that were reported at birth, with agreement highest for Pierre Robin syndrome and lowest for isolated cleft lip. The occasional use of "cleft lip" or "cleft palate" as generic terms for any type or combination of orofacial cleft may have contributed to some of the disparity shown in this table.

Age at death, as shown in table 2, was related to the reporting of clefts on death certificates. The highest proportion of unreported clefts was in children who were at least 1 year of age when they died. Infants who lived less than 1 day accounted for the next highest proportion of unreported clefts. Overwhelming conditions at birth, not directly related to the presence of the cleft, were frequent causes of death among children under 1 day of age. Causes of death among the older children were more frequently accidents and chronic diseases that were also unrelated to the presence of the cleft. These factors may account for the lower proportions of reported clefts on death certificates in age groups at the two extremes.

Table 3 shows the differences in reporting other congenital malformations on birth and death certificates of children born with clefts, according to type of malformation. Not surprisingly, external malformations were reported more frequently on birth certificates, and internal malformations were reported more frequently on death certificates. The large number of unspecified malformations reported on death certificates serves as a reminder that death certificates cannot be equated with birth certificates in their value as source documents for ascertaining specific anomalies.

In studies based on various types of records, it has been observed that children born with clefts who had additional malformations have shown greater risk of death than children having only a cleft (*4–7*). As shown in table 4, the birth certificates of 1,087 of the 1,838 deceased children with clefts in the present study contained reports of one or more additional anomalies. By supplementing the information on birth certificates with congenital malformations mentioned on death certificates, an additional 355 children were found to have multiple malformations. Thus by using death certificates the percentage of deceased children with multiple malformations increased from 59 percent to 78 percent. The proportionate increase was fairly uniform for isolated cleft lip, combined cleft lip and palate, and isolated cleft palate. Pierre Robin syndrome exhibited a larger shift to multiple malformations than was observed

Table 2. Deaths with and without report of cleft on death certificate, by age at death, children born with clefts, 1956–65

Age at death	All deaths	Cleft reported		Cleft not reported	
		Number	Percent	Number	Percent
Total	1,838	737	40	1,101	60
Under 1 day	623	225	36	398	64
1–27 days	691	317	46	374	54
1–11 months	429	173	40	256	60
1 year or more	95	22	23	73	77

Table 3. Source of reported malformations other than clefts by type of malformation, 1,838 deceased children born with clefts, 1956–65

Type of malformation	Total malformations	Certificate source			
		Birth only	Birth and death	Death only, number	Death only, percent
All	3,228	1,200	744	1,284	40
External	1,892	1,052	528	312	17
Internal	927	108	191	628	68
Unspecified	409	40	25	344	84

Table 1. Type of cleft reported on birth certificate compared with type of cleft reported on death certificate, deceased children born with clefts, 1956–65

Type of cleft reported on birth certificate	Total	Type of cleft reported on death certificate					
		None reported	Total reported	Isolated cleft lip	Cleft lip and palate	Isolated cleft palate	Pierre Robin syndrome
All clefts	1,838	1,101	737	62	338	271	66
Isolated cleft lip	239	160	79	49	18	10	2
Cleft lip and palate	800	444	356	12	282	61	1
Isolated cleft palate	740	478	262	1	38	194	29
Pierre Robin syndrome	59	19	40	.....	.....	6	34

Table 4. Deaths showing single and multiple malformations reported by type of cleft, children born with clefts, 1956–65

Type of cleft	Total deaths	At birth [1]			At death [2]		
		Number with cleft only	Number with multiple malformations	Percent with multiple malformations	Number with cleft only	Number with multiple malformations	Percent with multiple malformations
All clefts.....	1,838	751	1,087	59	396	1,442	78
Isolated cleft lip.....	239	120	119	50	62	177	74
Cleft lip and palate.....	800	343	457	57	186	614	77
Isolated cleft palate.....	740	252	488	66	132	608	82
Pierre Robin syndrome....	59	36	23	39	16	43	73

[1] Reported on birth certificates only.
[2] Reported on birth or death certificate or on both.

for the other types of cleft, but the number of cases was small.

Clefts seem to occur in association with a broad spectrum of other congenital anomalies, and the study of specific types may yield significant etiological clues. This study has demonstrated that neither birth nor death certificates alone give a true accounting of the number or types of other congenital malformations present in a child born with a cleft lip or palate. When investigation is limited to deceased children with clefts, matched birth and death certificates seem to be a useful tool in studying the associated malformations. For studying multiple malformations in surviving children with clefts, other sources will usually be necessary to ascertain the associated malformations.

REFERENCES

(1) Hay, S.: Variables in under-reporting of clefts on birth certificates. Public Health Rep 82: 985–993, November 1967.

(2) Hay, S., and Mackeprang, M.: Cleft lip and palate mortality study. Cleft Palate J 9: 51–63, January 1972.

(3) Hay, S., and Tonascia, S.: A classification of congenital malformations. Dental Health Center, U.S. Public Health Service, San Francisco, 1968.

(4) Division of Vital Statistics: Registry for handicapped children and adults: Annual report. Special Report No. 87. Department of Health Services and Hospital Insurance. British Columbia, 1964.

(5) Ivy, R. H.: Congenital anomalies as recorded on birth certificates in the division of vital statistics of the Pennsylvania Department of Health for the period 1951–1955, inclusive. Plast Reconstr Surg 20: 400–411, November 1957.

(6) Loretz, W., Westmoreland, W. W., and Richards, L. F.: A study of cleft lip and cleft palate births in California, 1955. Am J Public Health 51: 873–877, June 1961.

(7) McKeown, T., and Record, R. G.: Malformations in a population observed for five years after birth. *In* Ciba Foundation symposium on congenital malformations, edited by G. E. W. Wolstenholm and C. M. O'Conner. Little, Brown & Co., Boston, 1960, pp. 2–16.

MACKEPRANG, MURIEL (Dental Health Center, San Francisco), and HAY, SYLVIA: *Death certificates as supplementary sources of information in epidemiologic studies of clefts. Health Services Reports, Vol. 87, October 1972, pp. 759–761.*

Matched birth and death certificates of 1,838 deceased children whose birth certificates recorded a cleft of the lip or palate were studied. Clefts were reported on 40 percent of the death certificates, but only three-fourths of the reported clefts agreed with the type of cleft reported at birth. Fewer clefts were reported on death certificates of children who died under 1 day or over 1 year of age than of children who died 1 day to 11 months after birth.

For congenital malformations associated with clefts, external malformations were reported more frequently on birth certificates than on death certificates, whereas the opposite was true for internal malformations. At birth, 59 percent of the children were reported to have multiple malformations. With additional knowledge gained from the death certificates, the proportion of deceased children with clefts having associated malformations rose to 78 percent.