

Background and Methodology of a Study of Congenital Malformations

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TO WHAT EXTENT are the frequently observed variations in incidence of congenital malformations real, and to what extent do they reflect selective underreporting, misdiagnosis, and bias in the source documents and in the methods of investigation? These questions were of fundamental importance to investigators of the Division of Dental Health, Bureau of Health Professions Education and Manpower Training, who, in the National Cleft Lip and Palate Intelligence Service (NIS), use birth certificates for epidemiologic studies of clefts and other congenital malformations.

To investigate the dimensions of the problem, the Epidemiology Branch of the Division of Dental Health and the Division of Vital Statistics of the National Center for Health Statistics jointly undertook a project to assess the completeness and accuracy of information about congenital malformations and other items on birth certificates. The National Center for Health Statistics recognized the value of this study as a potential aid in planning national

tabulations and in improving the registration of vital statistics. This paper reports the background of the study and the methods used in conducting it. Results of the investigation itself will be published subsequently.

To evaluate the information on birth certificates, these documents were compared with hospital records. Hospital records are convenient source documents for comparison, and they are presumably more accurate and complete than birth certificates.

Background

In previous comparisons of hospital records with birth certificates, the completeness of malformation reporting on certificates was estimated to range from a low of 14 percent to a high of 80 percent (1-8). Each of these studies was limited either to an investigation of a selected number of malformations or to a small number of hospitals. In this study we planned to compare reports of all congenital malformations noted on hospital records with the birth certificates of infants born during 1 year (1963) in one State (Iowa).

Iowa was selected because a relatively high incidence of clefts and other malformations had been reported in the NIS, which probably indicated less underreporting than in the country as a whole. Moreover, Iowa's population was large enough to provide an adequate number of cases for study, yet not so large as to preclude

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reviewing all hospital births occurring in 1 year. Almost all births in that State occur in hospitals.

The health agencies and facilities of Iowa, both public and private, that participated in the study gave their full cooperation. The Iowa State Department of Health supported and assisted us throughout the study. After the objectives of the study were explained to their various officials, the Iowa Hospital Association, the Iowa Medical Society, and the Iowa Osteopathic Society gave the proposed study the endorsement and assistance necessary to initiate and complete it.

Medical records of 144 hospitals were studied. The statistical laboratory of Iowa State University was awarded a research contract to carry out the field operations, consisting primarily of reviewing hospital records of newborns and abstracting verbatim all references to congenital malformations.

A choice of procedures was offered to hospital administrators. One procedure permitted hospital personnel to review and abstract their own records; the other called for the field staff of the statistical laboratory to review and abstract the records. Under this dual system we soon saw that there would be confusion and possible non-comparability of results. Accordingly, a series of personal visits by staff members of the Division of Dental Health, the statistical laboratory, and the Iowa State Department of Health was made to suggest that an abstractor who was not in the hospital employ, but who was especially trained to the requirements of the study, be used to abstract the records. The administrators of the hospitals were assured that confidentiality of records would be maintained. All but two hospital administrators accepted this proposal. The two exceptions were hospitals in which approximately 800 and 2,000 live births occurred. At these two hospitals the medical records librarian took the responsibility for reviewing and abstracting the records, and the work was carefully monitored by the statistical laboratory's fieldwork supervisor. The records of both hospitals were retained in the study.

Interviewers employed by the staff of the statistical laboratory were given a comprehensive orientation and training program. They were given a list of common conditions of the

newborn that were not considered by the investigators to be congenital malformations. The workers were instructed to record any possible anomaly not specifically excluded by the list, thus reducing the possibility of not recording true malformations.

In 1963, a total of 58,583 live births were registered in Iowa. As shown in the following list, 58,243 babies, or 99.4 percent of all babies born in Iowa, were born in hospitals participating in this study. Records were reviewed for 57,909 births, constituting 99.4 percent of infants born in these hospitals. Thus the hospital records of 98.8 percent of all children born in Iowa in 1963 were reviewed for this study.

<i>Birth records</i>	<i>Number</i>
Infants born in participating hospitals:	
Hospital records reviewed.....	57,909
Hospital records abstracted.....	7,299
Infants not malformed.....	1,828
Infants malformed.....	5,471
Hospital records not abstracted.....	50,610
Hospital records not reviewed.....	334
Record not found.....	283
Record not listed.....	51
Not born in participating hospitals:	
Born in other hospitals.....	209
Born at home or in physician's office.....	131
Total	58,583

Method

The name, birth date, and medical record number of each infant born alive in each hospital were listed and a notation was made as to whether the infant's record was reviewed and, if reviewed, whether it was abstracted. The lists of newborn infants were transferred to magnetic tape and were matched by computer with the roster of live births registered in Iowa in 1963. This procedure resulted in an unmatched group of 674 registered births.

Originally we believed that all institutions in Iowa in which live births occurred had been included in the study, but it was discovered subsequently that five clinics and two maternity homes had been omitted. Of the 209 babies born in these facilities, 155 were born in one institution serving unwed mothers. Another 121 births occurred at home, and 10 occurred in physicians' offices. Records of 334 infants were not reviewed because they were missing from the hospital files when the abstracting was done, or they had never been listed in the hospital records.

The matching procedure also revealed 46 hospital records of births that had not been registered within the legally established time limit. With the assistance of the Iowa State Department of Health, some of these missing certificates were located among the files of delayed registration or under adoptive names. Registration of the remainder was undertaken.

A total of 7,299 records, or 12.6 percent of those reviewed, were abstracted onto specially designed forms. The abstracted information included selected medical and demographic data as well as verbatim statements referring to the malformations which would later be compared with corresponding items on the birth certificates. Because the abstractors were instructed to note any possible malformations, even those that were doubtful, 1,828 records were abstracted of infants who did not have bona fide malformations. Forceps marks, cranial moulding, and poor muscle tone are examples of these nonmalformations. Abstracted records of 5,471 babies were retained in the study.

Fetal Death Records

After the collection of data had begun for the comparison of records of live births, the investigators decided to enlarge the scope of the study by adding hospital records of all fetal deaths and fetal death certificates. The inclusion of these documents permitted a better estimate of the real incidence of congenital malformations in the outcome of all pregnancies than could be obtained by restricting the study to live births. Additionally we would be able to evaluate the completeness and accuracy of registration of fetal deaths, to evaluate the completeness and accuracy of reporting malformations and other variables on certificates of fetal death, and to estimate the magnitude of fetal wastage in the population under study.

Except for two basic differences, the portion of the study on fetal deaths followed much the same pattern as that employed in the collection and processing of data on live births. One difference was that hospital records of all fetal losses, not only those with congenital malformations, were abstracted. The other difference was that all fetal losses occurring in hospitals, regardless of length of gestation, were included in the study, although the comparisons of hos-

pital records with fetal death certificates were limited to those of more than 20 weeks' gestation because registration of earlier fetal losses is not required in Iowa. A total of 5,092 hospital records of fetal deaths were abstracted, 777 of which were determined to be of fetuses of more than 20 weeks' gestation. There were 733 certificates of fetal death.

In addition to the nonregistration of some fetal deaths of more than 20 weeks' gestation, certain irregularities in the recording of fetal deaths by the hospitals were discovered. For example, seven infants were classified by the hospitals as stillborn, although they were born alive and lived 1 day or less. Four of these were registered on fetal death certificates, and the other three were not registered at all. Other fetal deaths were recorded by the hospitals as live births. With time and patience these problems were resolved and the events were reclassified into their appropriate groups.

Each hospital abstract of live birth or fetal death was reviewed carefully, and malformations were coded by experienced personnel. To more effectively compare the malformations noted on the hospital records and on the birth or fetal death certificates, "A Classification of Congenital Malformations" (9), developed for use in the NIS, was expanded from a two-digit code to include a third digit for specificity of site, type, or other detail.

Neither the seventh revision of the "International Classification of Diseases" (ICD), in use at the time of the initial coding, nor the eighth revision with its improved section on congenital malformations was suitable for the detailed analysis desired by the investigators. Interested researchers, however, will discover that the categories of malformation in this study can be translated into the ICD categories without difficulty.

Pediatric consultants assisted in classifying malformations according to the ease by which each malformation could be identified at birth and by the severity of the defect. Malformations that are incapacitating or fatal were considered to be major. Other malformations judged to be less serious and often correctable were classified as minor. Insignificant malformations were deviations from the normal that were not considered to have medical significance.

An infant could have more than one congenital malformation designated on the hospital record. On 5,471 records of live births, 6,516 congenital malformations were recorded. Among 128 deaths of malformed fetuses, 201 congenital malformations were recorded. The three grades of congenital malformations—major, minor, or insignificant—are shown in the table. The classification for insignificant anomalies, which included conditions such as tongue tie, phimosis, and small pigmented nevi, will be excluded from most of the analyses to be presented in subsequent reports in this series.

Limitations

Although we hope this study will add substantially to our knowledge of the completeness and accuracy of reporting congenital malformations and other items on certificates of live birth and fetal death, serious limitations of the source material must be recognized. The reporting of congenital malformations depends upon the diagnostic skill of the physician who examines the child, his care in recording abnormal conditions on the hospital record, and the subsequent transfer of the notes from the hospital record onto the birth or death certificate.

Some physicians, through training or experience, tend to be more alert to certain types of anomalies than to others. Examination procedures of newborns vary at different hospitals. Many malformations, unless immediately fatal, are not detected or properly diagnosed until sometime after the hebdomadal period, during which time most birth certificates are filed. Some physicians will diagnose as malformations conditions that other physicians will consider within normal limits. The consultants to the study demonstrated that interpretation of these conditions varies widely.

Another problem occurs in transferring clinical notes to the birth certificates, because there is space for little more than a general reference to a gross condition. The certificate of fetal death used in Iowa in 1963 had no provision for noting conditions unrelated to the cause of death. Occasionally, there is some reluctance about entering information on the birth certificate since it will pass from the confidentiality of the medical record into the realm of public record. The training and motivation of the per-

Congenital malformations reported on hospital records, according to severity, Iowa, 1963

Severity of malformation	Live births		Fetal deaths	
	Number	Percent	Number	Percent
Total.....	6,516	100	201	100
Major.....	1,127	17	170	85
Minor.....	2,442	38	30	15
Insignificant.....	2,947	45	1

son who actually transcribes information on malformations from the medical record to the birth or fetal death certificate may be a significant source of variation in the completeness and accuracy of the data. Also, the local vital statistics registrar may be conscientious and query an incomplete record, or he may unquestioningly accept all certificates. Underlying all these problems are probably inadequacies in the recordkeeping systems of hospitals in which the births and fetal deaths occur.

Another possible limiting factor of this study was the completeness and accuracy with which the hospital records were reviewed and abstracted. We do not know how many records that described malformed babies were missed or were inadequately reviewed. We do not believe that many malformations were missed, because of the large proportion of birth records that were abstracted (12.6 percent). Also, the abstractors were conscientious and highly motivated workers who received thorough and sustained training and supervision. Unavoidably, circumstances within some hospital settings did occasionally produce situations in which it was difficult to pursue missing or incomplete medical records.

The portion of this study on fetal deaths has other limitations in addition to those already mentioned. Only those events happening in or coming to the attention of hospitals participating in the study are included in our data. Fetal losses early in pregnancy are frequently not even recognized as such, and many others do not result in hospitalization.

The results of the study will be published serially and will include analyses of completeness and accuracy of malformation reporting

on birth certificates, characteristics of hospitals related to completeness and accuracy of congenital malformation reporting on birth certificates, incidence of malformations in live births and fetal deaths, completeness and accuracy of reporting demographic and medical information (other than malformations) on certificates of live births, incidence of fetal deaths, and completeness and accuracy of fetal death registration.

Summary

An evaluation of the completeness and accuracy of congenital malformations and other items reported on birth certificates was undertaken jointly by the Epidemiology Branch of the Division of Dental Health, Bureau of Health Professions Education and Manpower Training, and the Division of Vital Statistics, National Center for Health Statistics.

All congenital malformations noted on hospital records of infants born in Iowa in 1963 were compared with birth certificates of those infants. Medical records of infants born in 144 hospitals were abstracted by staff of the statistical laboratory of Iowa State University. Records were found and reviewed for 57,909 infants, 98.8 percent of the 58,583 births registered in Iowa in 1963.

Hospital records were matched with birth certificates. Unmatched certificates represented misfiled records or births not occurring in participating hospitals.

Records of all 5,092 fetal deaths occurring in hospitals and 733 fetal death certificates were also included in the study for better assessment of total outcome of pregnancies.

Malformations were classified by three-digit codes and by severity and ease of recognition.

A total of 6,516 malformations were discovered among records of 5,471 live births and 201 malformations among records of 128 fetal deaths.

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