Birth Variables and the Incidence of Cleft Palate: Part II

RICHARD F. DONAHUE, PH.D

Denver, Colorado

It was the purpose of this research to study in a very large human population some of the variables which may be related to incidence of cleft lip and/or palate. This paper is the second of a series of reports regarding that investigation.

Method

The investigation was described previously (3) and was based on 5,838,855 birth records obtained from 17 state departments of vital records and statistics. Information for these records was obtained, in most cases, from the attendant at the birth who was responsible for the accuracy of the data on the birth certificate. From the 5,838,855 records, 6,070 infants with cleft lip and/or palate were identified. A control group was arbitrarily selected by taking the record fifth in order after the cleft. The data were stored and anlyzed by electronic computer systems. States were selected for the investigation on the basis, generally, of whether or not they noted cleft palate specifically on the birth record and, in the same way, the number of years surveyed in each state was determined by how long such information had been reported in that state.

Variables for Study

A total of 18 variables were selected. They were arbitrarily divided into three groups, generally depending upon the apparent temporal influences important in their determination: a) variables determined at conception (sex, color, plurality, maternal age, paternal age, birth order, and maternal nativity), b) variables related to gestation (length of pregnancy, associated anomalies, classification of cleft, complications of pregnancy, and prenatal care), and c) variables related to birth (geographical location, urban-rural location, legitimacy, month, weight, and attendant). This report concerns data for the second group of factors.

Results and Discussion

GESTATION. In this study, an infant was considered premature if it resulted from a pregnancy of less than 37 completed weeks. The question

Dr. Donahue is associated with the University Park Psychological Center, Denver.

| avoub | weeks in gestation period | | | | |
|--------------|---------------------------|-------|-------|-------------|---------|
| group | 30 and under | 31–35 | 36-40 | 40 and over | unknown |
| control | | | | | ····· |
| Ν | 23 | 168 | 3,580 | 491 | 122 |
| % | 0.5 | 3.8 | 81.7 | 11.2 | 2.8 |
| cleft | | | | | |
| \mathbf{N} | 55 | 171 | 3,561 | 498 | 99 |
| % | 1.3 | 3.9 | 81.2 | 11.3 | 2.3 |

TABLE 1. Gestation period for control and cleft groups, in number and %.

of whether prematurity has a stronger relationship to the length of pregnancy or to the birth weight is of interest in considering birth defects in general. As shown in Table 1, 5.2% of the cleft palate births in the sample were premature, while 4.3% of the control group were born before the thirty-sixth week of pregnancy. The only statistically significant difference in gestation range was found to be in the under-30-week category, in which 1.3% of the cleft group was born prematurely as compared with 0.5% of the control.

Although DeVoss (2) found no significant relationship between cleft palate and gestation period, Shapiro (16), Stiegler and Barry (18), Murphy (10), and Lutz (8), have found that cleft palate children tend to be premature. Kraus and his associates (7) presented data on an aborted fetal population, comparing it to the more usually studied fetal populations surviving birth. The prevalence of cleft lip and/or palate was 1.88% (or 19 per 1,000 abortions) in the prenatal age range 6 to 19 weeks. This is to be compared with a rate of 1.6 per 1,000 live births. They found the age distributions of cleft and total aborted specimens to be significantly different. The mean prenatal age at which the abortion occurred was 9.5 weeks for the cleft group and 13.0 weeks for the total sample.

Phair (14) found that 10% of the children in her cleft palate sample were born prematurely. Of the 4,285 birth records noting the length of pregnancy in the present study, 23 in the control group and 55 in the experimental group recorded less than 31 weeks of gestation, a difference between the groups of 0.8% of total births.

These results indicate that there is a difference between the number of cleft palate children and the number of control children who were born before 31 weeks of pregnancy. The significance of the difference (Kolmolgorov-Smirnov Test: $\chi^2 = .02992$) is difficult to evaluate because that category is only one of five considered in this study. (Table 1)

Associated Anomalies. Two states, Washington and Vermont, recorded associated anomalies on their birth records. As shown in Table 2, 93 (10%) of the cleft palate births had associated anomalies while 31

236 Donahue

| | associated anomalies | | |
|---------|----------------------|---------|--|
| group – | with | without | |
| control | 31 | 897 | |
| 70 | 3.3 | 96.7 | |
| cleft | 93 | 835 | |
| N | 10.0 | 90.0 | |
| % | | | |

TABLE 2. Associated anomalies for control and cleft groups, in number and %.

(3.3%) of the noncleft palate births had congenital deformities, a difference of 6.7%. The experimental group consisted of 835 births (96.7%) with no associated malformations, while the control group included 897 children born without defects.

Holdsworth (6) reported a 20% incidence of multiple anomalies in children with clefts. Lutz (8), Nichols (10), Vaughn (19), Fogh-Anderson (5), and Oldfield (12) have suggested that there is a high incidence of other abnormalities with cleft palate.

Beder and associates (1) found 14.5% of his cleft sample had associated anomalies, while Lutz and Moor (9) reported that 25% of their cases had deformities. Glover, in Lutz (8), found oral clefts associated with a variety of deformities and Peer and associates (13) found a relationship with hearing loss. Kraus and associates (7), in their aborted sample, found more than half (61.7%) of the 60 cleft specimens with associated malformations. The highest incidence of associated malformations reported in the literature is 25% (9).

Malformations found co-existing with oral clefts by Kraus and associates (7) were brachydactyly, syndactyly, club hands, club feet, imperforate anus, absence of genitals, and various skeletal dysplasias.

CLASSIFICATION OF CLEFT. Data regarding extent of the cleft were obtained from six of the states, based on 2,256 individuals. Shown in Table 3, the present data approximate the traditional 25% for cleft lip, 25% for cleft palate, and 50% for cleft lip and palate. This distribution is comparable to that reported by Veau, in Murphy (10), but differs considerably from those reported by both Oldfield (12) and Fogh-Anderson (5). Kraus and associates (7) found that 63% of their aborted sample had isolated cleft palate, 27% had combined lip and palate, and only 10% had isolated cleft lip.

REPORTED COMPLICATIONS DURING PREGNANCY. Five states included data about reported complications during pregnancy on their birth records, reported in Table 4.

Although it is impossible to know from the data specifically what the reported "complications" were, items such as diabetes, extrusions, and

| | | extent of cleft | | |
|---------------------|-------|-----------------|----------|-------------------|
| sample | total | palate only | lip only | lip and palate |
| present study | | | - | |
| N | 2,256 | 588 | 595 | 1,073 |
| % | 100 | 26.0 | 26.3 | 47.7 |
| Oldfield (12) | | | | |
| N | 1,041 | 358 | 233 | 450 |
| % | 100 | 34.4 | 22.4 | 43.2 |
| Fogh-Anderson (5) | | | | |
| Ň | 2,880 | 663 | 928 | 1,289 |
| % | 100 | 23.0 | 32.2 | 44.8 |

TABLE 3. Extent of cleft in present study and in Oldfield's (12) and Fogh-Anderson's (5) reports.

TABLE 4. Reported complications of pregnancy for control and cleft groups, in number and %.

| group | total | reported co | reported complications | | |
|---------|-------|-------------|--------------------------|--|--|
| | 10141 | yes | no | | |
| control | | | | | |
| N | 1,464 | 139 | $\substack{1,325\\90.4}$ | | |
| % | 100 | 9.6 | 90.4 | | |
| cleft | | | | | |
| Ν | 1,464 | 202 | 1,262 | | |
| % | 100 | 13.8 | 86.2 | | |

cystitis were mentioned on the Wisconsin birth records. Most states, however, coded a simple yes or no.

Other variables listed as complications during pregnancy were stress, thyroid deficiency, metabolism, malnutrition, radiation, toxemia, anoxia, anaemia, spontaneous abortion, and vitamin deficiency.

Montana, New Mexico, Wisconsin, Michigan, and Pennsylvania supplied a total of 1,464 birth records for each of the two groups. The results indicate that 13.8% of the experimental group noted one or more complications of pregnancy as compared to 9.6% in the control group.

In the present research, there were 202 reported complications of pregnancy recorded in the experimental group and 139 reported complications in the control group, a difference of 4.2% of the total 1,464 birth records surveyed on this variable. That difference was statistically significant.

PRENATAL CARE. Three states supplied data on length of the period

238 Donahue

| group | St. 4.1.7 | trimester | | |
|---------|-----------|-----------|--------|-------|
| | total | first | second | third |
| control | | | | |
| Ν | 1,432 | 1,001 | 301 | 130 |
| % | 100 | 69.9 | 21.0 | 9.1 |
| cleft | | | | |
| Ν | 1,447 | 968 | 334 | 145 |
| % | 100 | 66.9 | 23.1 | 10.0 |

TABLE 5. Time of initial prenatal care for control and cleft groups, in number and %.

of prenatal consultation. Iowa, Pennsylvania, and Missouri accounted for 1,518 birth records which were broken down into three categories: a) those receiving prenatal counseling during the first trimester, b) those making their first prenatal visit to their doctor during the second trimester, and c) those expectant mothers who did not consult a physician until the last three months of their pregnancy or until the birth itself.

As shown in Table 5, 66.9% of the experimental group sought medical advice during the first trimester. In the control group, 69.9% of the expectant mothers made their first visit during this period. Those mothers in the cleft palate group receiving prenatal care during the second trimester of their pregnancies amounted to 23.1% and in the control group, 21%. Ten per cent of the women in the experimental group and 9.1% of the women in the control group received medical attention for the first time during the final trimester. None of the differences were significant.

Summary

A total of 5,838,855 birth records were obtained from 17 state departments of vital records and statistics. From the group, 6,070 infants with cleft lip and/or palate were identified. A control group was selected by taking the record fifth in order after the record for the cleft birth appeared. Comparisons between the two groups were made in this report for length of pregnancy, associated anomalies, classification of cleft, complications of pregnancy, and prenatal care. More of the infants with cleft palate were premature and had associated anomalies than were the normal infants. The obtained distribution of cleft-type was comparable to that reported by other investigators.

> reprints: Dr. Richard F. Donahue University Park Psychological Center 2343 East Evans Avenue Denver, Colorado 80210

References

- 1. BEDER, O. H., COE, H. E., BRAAFLADT, R. P., and HOULE, J. D., Factors associated with congenital cleft lip and cleft palate in the Pacific Northwest. Oral Surg, oral Med., oral Path., 9, 1267-1273, 1956.
- DEVoss, H., Study of the factors relative to the incidence of cleft palate births from 1945 to 1949 in San Bernadino county. Speech Monog., 19, 31-38, 1952.
- 3. DONAHUE, RICHARD R., Birth variables and the incidence of cleft palate: part I. Cleft Palate J., 2, 282-290, 1965.
- 4. DONAHUE, R. F., Etiology of cleft palate from birth records. Ph.D. Dissertation, University of Denver, 1961.
- 5. FOGH-ANDERSON, P., Inheritance of Harelip and Cleft Palate. Copenhagen: Arnold Busck Co., 1942.
- 6. HOLDSWORTH, W. G., Cleft Lip and Palate. London: Whitefriars, 1951.
- 7. KRAUS, B. S., KITAMURA, H., and OOF, T., Malformations associated with cleft lip and palate in human embryos and fetuses. *Amer. J. Obst. Gyn.*, 86, 321–328, 1963.
- 8. LUTZ, K. R., Study of the relationship of the occurrence of cleft palate and the presence of associated deformities and other factors. *Cleft Palate Bull.*, 9, 47-48, 1959.
- 9. LUTZ, K. R., and MOOR, F. B., Study of factors in the occurrence of cleft palate. J. speech hearing Dis., 20, 163-166, 1955.
- 10. MURPHY, D. P., Congenital Malformations. Philadelphia: Lippincott, 1947.
- NICHOLS, R. S., COFFEY, R. J., WEBSTER, R. C., RUSSELL, J. A., and QUIGLEY, L. F., JR., Association of velopharyngeal incompetency with other abnormalities. *Cleft Palate Bull.*, 8, 2-3, 1957.
- 12. OLDFIELD, M. C., Some observations on the cause and treatment of harelip and cleft palate based on treatment of 1,041 patients. Brit. J. Surg., 46, 311-321, 1959.
- PEER, L. A., STREAN, L. P., WALLSER, J. C., BERNHARD, W. G., and PECK, G. C., Induction of cleft palate in mice by cortisone and its reduction by vitamins. J. Internat. col. Surg., 30, 249-254, 1958.
- 14. PHAIR, GRETCHEN M., Wisconsin cleft palate program. J. speech Dis., 12, 288-293, 1947.
- 15. SCHWARTZ, R., Familial incidence of cleft palate. J. speech hearing Dis., 19, 71-76, 1954.
- 16. SHAPIRO, R. N., ET AL., Incidence of congenital anomalies discovered in the neonatal period. Amer. J. Surg., 96, 398-399, 1958.
- 17. SPRIESTERSBACH, D. C., Personal communication. Department of Speech Pathology and Audiology, State University of Iowa, 1960.
- STIEGLER, E. J., and BARRY, M. F., New look at the etiology of cleft palate based on a study of 164 family histories. *Plastic reconstr. Surg.*, 21, 52, 1958.
- VAUGHN, H. S., Congenital Cleft Lip, Cleft Palate, and Associated Nasal Deformities. Philadelphia: Lee & Febiger, 1940.