

Seasonality, Incidence, and Sex Distribution of Cleft Lip and Palate Births in Trent Region, 1973-1982

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The birth dates of 930 children born in the Trent Region 1973-1982 with a diagnosis corresponding to cleft palate, or cleft lip with or without cleft palate, was obtained from Hospital Activity Analysis. These data were analyzed to look at the incidence of clefting, sex distribution, and seasonal variation. The results obtained show a significant peak incidence of cleft palate births in August and September, with a low incidence in March to May. Cleft lip, with or without cleft palate, shows a different seasonal trend, with peak occurrence in December and January and a low frequency in May and June. These seasonal trends may act as a pointer to the environmental factors active in the multifactorial etiology of cleft lip and cleft palate.

KEY WORDS: *cleft lip, cleft palate, incidence, sex distribution, seasonality*

"To everything there is a season, and a time to every purpose under the heaven: a time to be born, and a time to die."

Ecclesiastes 3

There is a divergence of opinion regarding the seasonal variation in the incidence of cleft lip and palate. While there is evidence of an overall seasonal variation in birth numbers (Cowgill, 1966) and evidence of seasonality in the birth of children with neural tube defects (Dallaire et al, 1984; Elwood, 1975), the situation regarding cleft lip and palate is unclear. The clinical impression is that there is an increased incidence in the birth of children with cleft lip and palate during the colder months of the year, in the winter and spring. Several authors have found a seasonal incidence in cleft lip and palate births (Edwards, 1961; Fujino et al, 1963; OPCS, 1983; Sandahl, 1977; Saxen and Lahti, 1974, Wehrung and Hay, 1970); others however, have found no such variation (Chapman, 1983; Fraser and Calnan, 1961; Heath, 1977; Slater et al, 1964; Woolf et al, 1963).

The present study examines the seasonal variation in the birth date, the incidence and sex dis-

tribution in the birth of children with cleft palate or cleft lip with or without cleft palate within the Trent Region during the period 1973 to 1982. The Trent Region is situated in the East Midlands of England with a population of approximately 5,000,000 in mixed and rural communities.

METHOD

Hospital Activity Analysis (HAA) figures were used to obtain the birth dates of children with cleft lip, cleft palate, or both. Hospital Activity Analysis is produced from data obtained from the clinical notes of each patient discharged from hospital in England and Wales. This information is compiled by each Regional Health Authority to produce a comprehensive data bank giving such personal details as name, date of birth, and clinical details such as diagnosis and operations. An alternative source of information on cleft palate births is the congenital malformation scheme run by the Office of Population, Census and Surveys (OPCS). However, HAA data were considered to be more accurate than the congenital malformation scheme. Children with cleft anomalies appear on HAA because of admission either to a special care baby unit in the neonatal period or to a hospital for surgical repair of the

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cleft; the OPCS scheme relies on voluntary notification and only covers malformations identified within 7 days of birth. Permission was obtained from each hospital or doctor in the Trent Region under whose care a child had been admitted with a diagnosis-code corresponding to a cleft lip or palate anomaly (ICD codes 7490, 7491, 7492). This was required to eliminate multiple entries for the same patient since HAA is an event-based record rather than a patient-based record, and OPCS files data by patient name.

The patient's name, area of residence, sex, date of birth, and diagnosis were entered on a simple database program on a microcomputer. The personal data were searched and multiple entries deleted. The diagnostic data were examined under two categories; isolated cleft palate (ICD code 7490) and cleft lip, with or without cleft palate, (ICD code 7491, 7492) as these malformations are considered to be genetically distinct (Fogh-Andersen, 1942). The total number of entries for each of the three diagnoses for each month of the 10-year period was obtained and aggregated to give monthly totals. The monthly totals were adjusted to take account of the normal seasonal variation in number of births. Three monthly moving averages were used to remove the minor fluctuations in the data and thereby reveal any underlying trend.

RESULTS

A total of 930 children, born within the period 1973 to 1982 appeared in the HAA data for the Trent Region, with a diagnosis of cleft palate or cleft lip with or without cleft palate. Of these children, 364 (39 percent) had isolated cleft palates (CP), and 566 (61 percent) had cleft lip with or without cleft palate (CL(P)). In the CL(P) group, there was a higher incidence of males, 352 males (62 percent) and 214 females (38 percent). The isolated cleft palate group had a higher proportion of females, with 199 females (55 percent) and 165 males (45 percent).

The number of cleft palate births per year over the period studied is given in Table 1. The incidence per 1,000 live births is also shown, the overall incidence during the period studied was 0.70 per 1,000 live births (Table 1). Table 2 shows the number of CL(P) births and the incidence over the 10-year period. The overall incidence was 1.12 per 1,000 live births.

There would appear to be a reduction over time in the incidence of both cleft palate and cleft lip with or without cleft palate, but there are fluctuations between the years. Figures 1 and 2 show the aggregated number of births of children with isolated CP and with CL(P) per month, using 3-monthly moving averages. These have been

TABLE 1 Cleft Palate Births for Trent Region, 1973-1982

<i>Year</i>	<i>No. Of Births</i>	<i>Rate per 1,000 Live Births</i>
1973	37	—*
1974	45	0.87
1975	38	0.76
1976	36	0.73
1977	36	0.75
1978	38	0.75
1979	32	0.60
1980	41	0.73
1981	26	0.48
1982	35	0.66

* Regional birth data not available.

corrected for normal seasonal variation in the number of births in Trent Region. In the CP group, the number of births in the period Feb-Mar-Apr, Mar-Apr-May and Apr-May-Jun falls two standard deviations or more below the annual mean, whereas for the period Jul-Aug-Sep and Aug-Sep-Oct the number of children born with cleft palate is more than two standard deviations above the annual mean (Fig. 1). For the CL(P) group, the Apr-May-Jun and May-Jun-Jul period showed the number of births of children with clefts to be more than two standard deviations below the annual mean whereas for the period Nov-Dec-Jan and Dec-Jan-Feb the number of births of children with clefts was more than two standard deviations above the annual mean (Fig. 2).

DISCUSSION

In this sample of children with cleft lip and palate who were studied using the data available from the Trent Region, 36 percent of the overall number of cleft defects were isolated cleft palates and 64 percent were cleft lip with or without cleft palate. This distribution agrees with the figures found by Abyholm (1978) with respect to a Norwegian population and is in accord with the distribution quoted by Robertson (1983).

TABLE 2 Cleft Lip With or Without Cleft Palate Births for Trent Region 1973-1982

<i>Year</i>	<i>No. Of Births</i>	<i>Rate Per 1,000 Live Births</i>
1973	42	—*
1974	77	1.47
1975	56	1.11
1976	67	1.35
1977	47	0.97
1978	51	1.01
1979	59	1.11
1980	70	1.25
1981	49	0.90
1982	48	0.91

* Regional birth data not available.

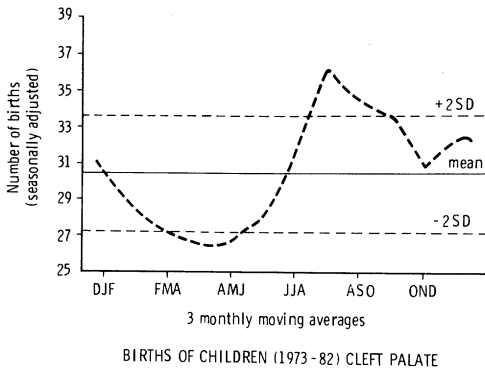


FIGURE 1 Incidence of children born with cleft palate by 3-monthly moving averages between 1973 and 1982.

The sex distribution in the study group shows a greater number of males in the CL(P) group, with 352 males and 214 females. This sex distribution is consistent with the findings of other workers looking at a predominantly Caucasian population (Abyholm, 1978; OPCS 1983; Saxen and Lahti, 1974; Woolf et al, 1963). The isolated cleft palate group had a higher female incidence, 199 females compared with 165 males; this agrees with other studies that also found CP to be more common in females (Abyholm, 1978; Bhatia, 1972; OPCS, 1983; Poole, 1975; Saxen and Lahti, 1974) with a reported ratio of 2:1 (Robertson, 1983). The ratio found in this present study in the Trent Region is lower at 1.2:1.

The overall incidence rate of isolated cleft palate births in the 10-year period studied is 0.70 per 1,000 live births. This incidence rate is similar to that found by Chapman (1983) in a study in Auckland, New Zealand, who reported 0.83 per 1,000, and Saxen and Lahti (1974) who found an incidence rate of 0.86 per 1,000 in Finland. Robertson (1983) quoted a lower overall incidence rate of 0.45 per 1,000. The OPCS notifications show an overall incidence rate of 0.46 per 1,000 in England and Wales, with an incidence rate in the East Midlands Standard Region of 0.57 per 1,000, which is the area of highest incidence in England and Wales. The incidence rate of CL(P) in the Trent group is 1.12 per 1,000; this is similar to that reported by Chapman (1983) who reported an incidence rate of 1.089 per 1,000 in Auckland. In Finland (Saxen and Lahti, 1974) a lower incidence rate of 0.83 per 1,000 was reported. The incidence found in the present study lies within the frequency range given by Robertson (1983) of 0.8 to 1.6 per 1,000. The overall incidence rate in England and Wales from OPCS notifications is 0.95 per 1,000, with an incidence rate in the East Mid-

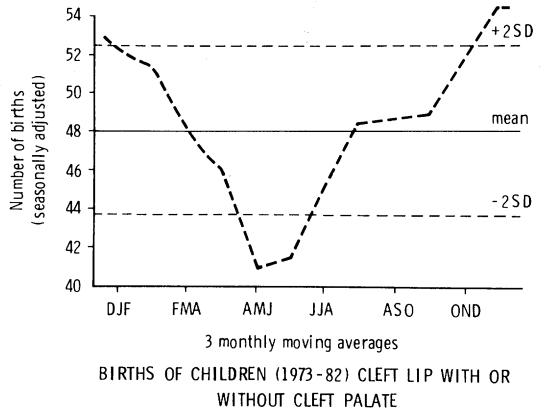


FIGURE 2 Incidence of births of children with cleft lip with or without cleft palate by 3-monthly moving averages between 1973 and 1982.

lands of 1.07 per 1,000. This would suggest that there has been a high level of identification of children with cleft deformities within the Trent Region by the method adopted in this study.

The variation in the incidence of CP births in this study with that found by the OPCS notification scheme could point to an apparent deficiency in the coverage of the notifications, or it may suggest that there is a much higher prevalence of CP births in the Trent Region compared with the rest of England and Wales.

The results also suggest that there is a seasonal incidence in the birth of children with CP and CL(P) anomalies but that the two differ. Seasonality with respect to the incidence of congenital anomalies is a well recognized phenomenon and has been reported in neural tube defects (Dallaire, 1984) when the date of conception was taken as the last menstrual period (LMP). Edwards (1961) reported a seasonal variation in the incidence of anencephaly with a higher occurrence in winter births. However, the situation regarding seasonality in cleft defects has been unclear. No seasonal variation was found in Utah by Woolf et al (1963) who studied the surgical records of 418 patients covering a 35-year period. Yet, Heath (1977), who looked at children born in the Oxford area during the period 1965 to 1974, found some evidence for significant clustering in space and time during the period 1965 to 1971, but this was not repeated when 1972 to 1974 data were studied. In the study on a Polynesian community in Auckland, Chapman (1983) found no significant trends, and no evidence of seasonal differences in incidence were reported. Other workers, however, have found seasonal variations in the incidence of cleft defects. Saxen and Lahti (1974) in Finland found no evidence when pooled cleft data were used, but when CL(P) was separated from CP and additional malformations were excluded, they

found a high incidence of CL(P) and a low incidence of CP in the last quarter of the year. Wehrung and Hay (1970) in a study in the United States reported that cleft lip with or without cleft palate, hypospadias, and positional foot defects all demonstrated statistically significant seasonal trends, with a high incidence in March. Edwards (1961) looking at the incidence in congenital disease in Birmingham found a seasonal trend in cleft lip but not in other clefts; this study, however, only looked at births during 1 year, 1950. Fujino et al, (1983) analyzed the data of 2,828 Japanese patients with cleft defects and found seasonal effects to be apparent, with CL(P) births decreased during winter months (Dec-Feb) and increased in spring (Mar-May). They found no significant trends for isolated cleft palate. Sandahl (1977) used data relating to LMP and found a peak incidence for CP was when the LMP occurred in April, i.e., births in February. In the 10-year period 1971-1980, the OPCS notification scheme shows a decrease in the CL(P) births in October, and a higher rate in December and January. No such seasonal effect was shown in CP births.

The seasonal trend found in this study in the Trent Region shows a reduced incidence of children born with isolated cleft palate in March to May, with increased incidence August to September. This, therefore, differs from the findings of Saxen and Lahti (1974) in Finland, who found a low incidence of CP births in October to December but noted a constant incidence throughout the rest of the year.

In the current study, the CL(P) group shows a different pattern through the year relative to the CP group, with a low incidence in May-June and a high incidence in Dec-Jan. Hence, the results are in keeping with those reported by Saxen and Lahti (1974) who divided the year into four quarters and also found the peak incidence to be at the end of the year, during the period Oct-Dec. In another study, Sandahl (1977) in Sweden also found a winter peak incidence for CL(P) births since they related to mothers whose LMP occurred in March, i.e., with respect to January births. However, in a Japanese population, Fujino et al (1963) found CL(P) significantly reduced in the spring (Mar-May) in contrast to Charlton (1966) in Australia, and Wehrung and Hay (1977) in the United States who reported a peak incidence of CL(P) in March.

There are various possible explanations to account for seasonal variations in congenital malformations. The presence of a teratogen at a specific time of the year, e.g., viral infections such as rubella, which can cause developmental

anomalies, tend to show seasonal trends. Alternatively, metabolic or endocrine factors may affect seasonal incidence, although recent research into this has found no evidence to support this contention (Neibyl et al, 1985). External environmental factors have been looked at; the use of agricultural chemicals was examined in relation to cleft deformities in Iowa and Michigan; however, once again no association was found (Gordon and Shy, 1981).

Dietary factors may be of importance. Experimentally, vitamin A and riboflavin deficiency in rats have been shown to induce a variety of malformations, including cleft palate, and high doses of vitamin A have been used to induce clefts in animal experiments (Shafer et al, 1983). It is therefore possible that seasonal variations in availability and, in consequence, intake of natural sources of vitamins, for example fresh fruit and vegetables, may play a part in the multifactorial etiology of clefting and other environmentally influenced congenital malformations.

The evidence presented in this descriptive study suggests that seasonal factors are involved in the development of CL(P) and CP. Since the peak incidence of isolated cleft palate in the Trent Region is August to September, this implies that the normal developmental process has been affected during the later part of the second month of development, i.e., January or February of the year in question. With respect to cleft lip with or without cleft palate, the peak incidence found in this study is December to January, i.e., the crucial period for possible environmental factors is May and June. The seasonal trends may act as a pointer to the identification of these factors.

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