Services for Children with Congenital Facial Clefts Through a State Crippled Children's Service Program

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A study to analyze the involvement of the Maryland State Crippled Children's Services (CCS) with children with congenital facial clefts was conducted in two parts. The first phase identifies all children born to Maryland residents over a three-year period to measure the incidence by type of the cleft condition. Phase two examines CCS involvement for 816 children with congenital facial clefts, including 114 of the Phase 1 cohort group. Diagnostic and demographic data are analyzed with specific attention to services involved in the child's entry into, flow through, and discharge from the CCS program.

The 816 study cases known to the Maryland CCS Program during the period of study were characterized by the presence and type of parental congenital malformations, reasons for termination of CCS services, number of professional encounters, age at entry, distances to CCS care providers, demographic characteristics, and hospitalization history.

Data analysis suggests that the age at which a child first becomes known to the CCS Program is significantly associated with areas of residence, presence of other malformations, type of cleft, and presence of other malformations in the family. The data further suggest that CCS Programs reassess their capacity to develop administratively useful data for use in program planning, evaluation, and research. The present systems appear to require further alteration for updating and expansion to increase the availability of useful, timely, and accurate programmatic data.

KEY WORDS: case finding, cleft lip, cleft palate, state crippled children's services

Introduction

A study was undertaken to examine questions related to the detection, diagnosis, treatment, and dispostion of cases involving children with congenital facial clefts relative to actual or potential involvement with the state Crippled Children's Services (CCS). The study included two phases. The first was directed at identifying a cohort of children born to Maryland resident mothers between 1968 and 1971 (n=289) to establish incidence and to determine the number of children eligible by clinical conditions for CCS services. The second phase was designed to examine the flow of children identified in the cohort study plus others known to the state CCS during the period of 1969 through 1976 (n=816). While incidence data are provided, this report is primarily directed at delineating medical, sociodemographic, and economic information relative to case identification and the provision of services for children born with congenital facial clefts.

Background

There have been numerous attempts to determine the incidence of congenital facial clefts in representative population samples (Greene, et al., 1965: Hay, 1971; Kakalik, 1973; Milham, 1963). Recent variations range from 1.84 (Emanuel, et al., 1973) to 1.28 (Hay, 1967) per 1,000 live births. Meskin (1966) provided a succinct summarization of studies over the last century and reported a range from .53 among World War I army draftees to 1.43 for a 1956–1964 state study based upon birth records. The Hay study (1967) provided data from 26 birth registration areas in the U.S. and examined differences between birth certificate registration and the records of Crippled Children's Ser-

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vices. She noted greatest birth certificate recordings where the lip and palate were both involved (71.4%) and lowest completion (61.2%) where only the palate was involved. Variations in reportability were also noted by geographic area, race, birthweight, and place of delivery.

Nearly a decade ago it was estimated that only about 6% of the handicapped children were being served through the combined efforts of the CCS programs (Kakilik, 1973). Appreciable variations reported in the services provided by state CCS programs and summary data likewise reveal differences between regions and within states. Although each state provides some reasonably unique orientation to Crippled Children's Services programs, there appears to be a commonality in focus, objectives, service mandates, and the nature of funding for such programs. The administrative focus of various state CCS programs differs, with 34 state programs located in state health agencies, four in university-based programs, nine in state welfare agencies, three in Departments of Human Services, and three in state education departments (Wallace, 1968). By federal regulation, the programs are directed by a physician.

Federal funds are allocated to each state under Title V of the Social Security Act, Section 504, with the allotment being determined by federal appropriations and a formula predicated on the financial needs and the number of crippled children in need of services within each state (Social Security Act, 1935).

Methodology

Ascertaining the total number and rate of children with congenital facial clefts required use of multiple data sources. Demographic data for 203,802 resident live births to Maryland mothers during 1969 through 1971 were examined through use of birth registrations, State Crippled Children's program registry, specialty hospital records, records of local health departments, discharge diagnoses following the child's birth. All Maryland resident births in Maryland, the District of Columbia, Delaware, and Pennsylvania hospitals were examined to produce the complete enumeration of the 289 cohort children. The single most effective source of cohort identification was the review of hospital birth information, which identified 279 (96.5%) of the cohort children compared to only 161 (55.6%) identified from birth registrations.

At different stages of the study, selected service utilization (encounter, distance, and provider) and financing variables were reexamined for 56 randomly selected cases. Data on the sources of financing were not significantly different ($r = .694$) among the multiple abstractions, whereas service utilization data reached considerably higher r values. With respect to cleft diagnostic types, classification was based on 90% or better agreement on repeated individual record entries. Where inconsistency was found, the composite records were examined to determine the most reasonable diagnostic category.

Once identification of all children with congenital facial clefts was made, these records were merged with State Crippled Children's records to determine the proportion of eligible children actually known to State CCS, the age at entry, age at discharge by cause, and the services received while in the program. An eight-year period (1969 through 1976) was examined to secure CCS data essential to assess adequately the entry, flow, services, and exit components of the program.

Information from the Central CCS Office included only those children receiving services from physicians, dentists, and hearing and speech pathologists. It excluded services provided through the local health departments by public health nurses, social workers, nutritionists, or other professionals. Therefore, data pertaining to other services were obtained by a review of all records of local health departments and specialty providers.

Results

Examination of the cohort data revealed 55 cases more than would have been expected using traditionally accepted rates of congenital facial clefts. Exacting procedures for enumerating the occurrence of congenital facial clefts and omitting duplicate counting among the cohort children precluded the possibility of overenumeration.

Of the 289 cohort children, 83.0% (n=240) were classified as Caucasian while 17% (n=49) were non-white children. The cohort children were represented in each of the cleft types

noted by Meskin (1966) with 20.8% having cleft lip only, 42.6% with cleft lip and palate, 35.6% with cleft palate only and 1.0% of the children with a submucosal cleft condition. See Table 1. The sex distribution is consistent with other reported findings. Subjects with bilateral cleft lip and cleft palate represent 17.3% of the total. This distribution for lip and palate is in agreement with that found by Meskin (1966), although the categorization by completeness of cleft varies significantly and suggests the definition and categorization of completeness differs between the studies. Within the cleft lip alone categories, the unilateral lip occurred most frequently with the current data suggesting no significant difference between the left or right side. Within the palate only category, the complete clefting or hard and soft palate occurred most frequently.

Other malformations were recorded for 72 cohort children (24.9%). A single additional malformation was recorded for 17 of the 72 (23.6%) and 48 children (66.7%) had five or more other malformations.

Table 2 reveals pregnancy associated characteristics for the cohort children. Over 88% were single births and 18.3% were low birth weight children. Prenatal care commenced in the first trimester for 61.6% of the mothers. Sixty-one percent of the mothers received their prenatal care from private physicians,

TABLE 1. Distribution of Facial Cleft by Type of Diagnosis

Type of Cleft Condition Description	*Cohort Children*		*CCS Known Children*		*Ratio Cohort: CCS*
	Number	*Percent*	*Number*	*Percent*	
A. *Cleft Lip*					
Complete Bilateral	3	1.0	10	1.2	1:3.3
Incomplete Bilateral	9	3.1	5	0.6	1:0.6
Complete Right Side	2	0.7	8	1.0	1:4.0
Incomplete Right Side	19	6.6	13	1.6	1:0.7
Complete Left Side	8	2.8	13	1.6	1:1.6
Incomplete Left Side	19	6.6	20	2.5	1:1.1
B. *Cleft Lip with Cleft Palate*					
Complete Bilateral	47	16.3	156	19.1	1:3.3
Incomplete Bilateral	3	1.0	12	1.5	1:4.0
Complete Right L/P	33	11.5	69	8.5	1:2.1
Incomplete Right L/P	4	1.4	9	1.1	1:2.3
Complete Left L/P	27	9.3	120	14.7	1:4.4
Incomplete Left L/P	9	3.1	15	1.8	1:1.7
C. *Cleft Palate*					
Complete of Hard and Soft Palate	35	12.1	131	16.1	1:3.7
Incomplete (hard) and Complete (soft) Palate	31	10.7	80	9.8	1:2.6
Complete of Soft Palate	23	8.0	59	7.2	1:2.6
Incomplete of Soft Palate	14	4.8	30	3.7	1:2.1
Palatal Shortness	—	—	9	1.1	—
Combination of Submucous and Regular Clefts of the Hard (Partial) and Soft (Complete) Palate	1	0.3	1	0.1	—
Combination of Submucous and Regular Clefts of the Soft Palate	2	0.7	37	4.5	1:1.0
Unverified Diagnosis	—	—	19	2.3	1:1.8
TOTAL	289	100.0	816	100.0	1:2.8

TABLE 2. Summary of Pregnancy Associated Characteristics of Cohort Children and Medical Assistance Status at Birth (n = 289)

	Number	*Percent*
A. Plurality of Birth		
Single birth	256	88.6
Twin-first born	8	2.8
Twin-second born	7	2.4
Triplet	1	0.3
Unknown	17	5.9
B. Birthweight		
<2000 gms	24	8.3
2001–2500 gms	29	10.0
2501–3000 gms	58	20.1
3001–3500 gms	77	26.6
>3500 gms	78	27.0
Unknown	23	8.0
C. Initiation of Prenatal Care		
First trimester	178	61.6
Second trimester	59	20.4
Third trimester	16	5.6
No prenatal care	5	1.7
Unknown	31	10.7
D. Source of Prenatal Care		
Private physician	177	61.3
Hospital clinic	68	23.5
Health Department clinic	13	4.5
Other	2	0.7
No prenatal care	5	1.7
Unknown	24	8.3
E. Medical Assistance (Title XIX) Status		
Certified	24	8.3
Not certified	228	78.9
Unknown	37	12.8

and only 8.3% certified for care under Title XIX (Medical Assistance).

Of the 289 cohort cases, 104 (36%) were actually known to the state CCS program office. These 104 cases were examined in conjunction with 712 additional CCS cases that were born in other than the cohort years. Data from the analysis of all 816 CCS known cases reveal that 589 (72.2%) were receiving clinical services through arrangements by the Central CCS office. An additional 8.3% were known to the Central Office but had not received direct services, and 15.6% were receiving services only from the local health department programs. An additonal 3.9% were receiving care and services through local health departments and specialty hospitals combined.

Over the eight-year period covered by this phase of the study, 632 of the children (77.4%) had continued care under the Crippled Children's program. Reasons cited for termination of CCS cases suggested that the most frequent involved out-of-state moves by the family. The continuity of service, described as a primary objective of state CCS programs, was complemented by the high continuation rate but raises questions concerning efficiency and efficacy.

An analysis of geographic area between the birth cohort and the cases known to CCS suggests that becoming known to a CCS program is accomplished through multiple factors including geography. A general trend was seen for the more rural areas and inner-city areas to have a higher ratio of cohort children known to the Crippled Children's program. *While no definitive explanation is present, it can be speculated that a central city population might tend to be somewhat less financially able to pay costs for the expensive care involved in treating a complex problem like facial clefts.* People in rural areas might experience economic problems and shortages of professional resources necessitating referral. Furthermore, nearly 75% of the Baltimore City resident births occur in the four teaching hospitals where staff may be inclined to refer cleft treatment to the local specialty hospitals which provide care in conjunction with the Crippled Children's Service program. The non-urban areas not only lack specialty hospitals but also lack private practitioner specialty resources necessary for the early detection and referral for specialty care. The combined impact of availability of medical care specialty resources and economics are likely strong reasons for CCS referral.

Cases known to the state CCS program have a similar diagnostic profile to the cohort children (Table 1). The diagnosis of combined lip and palate involvement accounted for the largest single group of CCS cases (46.7%) as compared to 42.6% for the cohort children. However, when comparing cohort and CCS cases with duplicate cases removed, the combined lip and palate group is significantly over-represented whereas the lip only or palate only groups are less represented in the CCS caseload.

Excluding the 19 cases with unverified diagnoses in the CCS group and using an unduplicated count, the difference between what would be expected and what existed by diagnostic group was highly significant ($x_3^2 = 58.6$, $p < .001$). Whereas only 23.8% of the

unduplicated cases in the cohort group had cleft lips, a considerably smaller proportion (7.4%) of the CCS caseload group had that diagnosis. Conversely, while 29.2% of the cohort children carried a diagnosis of lip and palatal involvement combined, 46.6% of the CCS caseload children were so involved. The data suggest that youngsters whose clefts were confined to the lip or palate only were provided with less CCS subsidized care, possibly because their needs were not so great and the expense was lower than would be the case if the deformity were more extensive. The financing systems, which did not record linkage with Medical Assistance (Title XIX), raised a question as to whether such systems showed the financial need or a perceived need of a diagnostician or service coordinator for the more comprehensive and continuous care provided under a state CCS program.

Although one might expect early identification and referral to a Crippled Children's program, data revealed that only 2.3% of the 816 children received their first CCS service by the age of 12 months. Only 21.7% had been enrolled in CCS by the end of their fifth year, and 15.9% of the 816 children received their first CCS service between the ages of 16 and 20 years (Table 3).

Data revealed that the 816 children had a total of 15,552 face-to-face encounters within the State CCS program (mean = 19.1). Although 35.7% of the children (n=291) had fewer than five encounters, 23.3% had more than 30 encounters.

Over one-third (37.4%) of the 816 children had no recorded hospitalizatons for facial clefting under this CCS program. The remaining 511 children averaged 38.1 hospital days during their CCS coverage.

The State CCS care was provided at various satellite locations throughout the political subdivisions and at two specialty hospitals located in Baltimore City. Various plastic, dental, speech and hearing, and other services were also available in private facilities within and contiguous to the state. Table 4 reveals that 34.9% of the children reside within the immediate area of the facility from which service was provided. In some cases, the family resided very close to the service-providing facility, but 190 of the families (23.3%) traveled a mean one-way distance in excess of 20 miles to receive treatment. Further analysis of the data revealed that the outpatient plastic, hearing, and speech services were more frequently within the same community, whereas other specialty programs and inpatient care required traveling greater distances. The mean one-way distance traveled, computed from the mean of all known distances, was 14.4 miles (Table 4).

TABLE 3. Age of Patients with Facial Clefts When First Enrolled in the CCS Program

Age	*Number*	*Percent*	*Cumulative Percent*
<1 year	19	2.3	2.3
1–5 years	158	19.4	21.7
6–10 years	213	26.1	47.8
11–15 years	233	28.6	76.4
16–20 years	130	15.9	92.3
>20 years	63	7.7	100.0
TOTAL	816	100.0	

TABLE 4. Mean One Way Distance Traveled by CCS Children for Facial Cleft Treatment

Mean Distance	*Number*	*Percent*	*Cumulative Percent*
Within same community	285	34.9	34.9
2–5 miles	83	10.2	45.1
6–10 miles	76	9.3	54.4
11–15 miles	89	10.9	65.3
16–20 miles	59	7.2	72.5
21–30 miles	109	13.4	85.9
31–40 miles	53	6.5	92.4
41–50 miles	28	3.4	95.8
51–171 miles	816	100.0	
TOTAL			

Sum of one way distance traveled = 11,753
Mean Distance = 14.4 miles

Discussion

The usually higher occurrence of low birthweight among the cohort children mandates further examination of those factors which may be associated with birthweight and congenital facial clefts. Prior literature has not identified a difference in birthweight except when multiple handicapping conditions were present. Fifty-three of the 289 children were less than 2500 grams at birth. The examination of the physical and psychosocial factors involved in those cases must be used to elicit further information since the low birthweight child begins life under other than optimal circumstances.

Facial clefts were selected as a prototype condition for examination because of several factors, not the least of which was the presumed early and complete identification of the condition. If only hospitalization discharge data were used, 96.5% of the cohort children could have been identified. Although these data are not routinely utilized by a state CCS Director, use of hospital data could effectively enhance early detection and entry into treatment. However, two basic questions arise. (1) Is early intervention by a CCS program necessary to prevent secondary physical, social, language, or emotional problems? (2) What proportion of the children can significantly benefit from services through a state CCS program? Data on the age of entry suggest that more than half of those ultimately known to CCS were still unknown by the age of ten years. It would be presumed that physical correction had been in process from a much earlier time, but provision of associated services to prevent secondary problems is not documented. Furthermore, if the CCS program is for more than economic support, what happens to the 64% who never become known? Do they receive timely and comprehensive restorative services?

The referral to CCS, according to the current data, is dependent upon a combination of factors. In part, the CCS is a financing mechanism for those who are above scale for Medicaid (Title XIX) funding but still without sufficient independent or third-party resources. The data go much further and suggest that referral to a CCS program is not solely on a financial basis but is also prompted by the multiplicity of services deemed necessary, differences in geographic area, cleft type, and distance to treatment facilities. Variations among states would be expected based upon individual differences in case finding and techniques, availability of specialty resources, financing agreements, and program goals.

The impact of comprehensive, continuous, and collaborative services provided by CCS programs, private physicians, and other professionals in the health field was not the focus of this study. One can wonder where those children, regardless of financial resources, would receive their needed specialized medical, surgical, and other care in the absence of specialists supported and encouraged by CCS programs. Further, the necessary supportive services are frequently unavailable except through local health department programs for handicapped children. Even in the more urban areas where specialists tend to congregate, the access to the multiplicity of service providers is questionable.

Service programs which do not include a deliberate effort directed toward data management lose the ability to base decisions upon easily accesible data. Administrative decisions appear to rely upon clinical judgment which must be recognized as lacking the precision believed necessary for making some complex decisions regarding service eligibility, program coverage, and allocation of program resources. The administrator must determine what precise information is essential for program management and the means by which such information can be made readily available. Organization of data systems and information in an administratively useful format is predicated on the administrative perception of its needs which, in turn, is based upon the availability of timely and accurate data.

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References

Emanuel, I.; Culver, B. H.; Erickson, J. D.; Guthrie, B.; and Schuldberg, D., The further epidemiological differentiation of cleft lip and palate: A population study of clefts in King County, Washington, 1956–65, *Teratology, 7,* 271–282, 1973.

Greene, J. C.; Vermillion, J. R.; and Hay, S., Utilization of birth certificates in epidemiologic studies of cleft lip and palate, *Cleft Palate J., 2,* 141–156, 1965.

Hay, S., Variables in underreporting of clefts on birth certificates, *Public Health Reports, 82,* 985–993, 1967.

Hay, S., Incidence of selected congenital malformations in Iowa, *Am. J. Epidemiol., 94,* 572–584, 1971.

Kakalik, J. S.; Brewer, G. D.; Doughtery, L. A.; Fleischauer, P. D.; and Genensky, S. M., Services for handicapped youth: A program overview, *Rand,* R1220-HEW, May, 1973.

Meskin, L. H., An epidemiologic study of factors related to the extent of facial clefts. (Unpublished thesis, University of Minnesota, 1966).

Milhan, S., Underreporting of incidence of cleft lip and cleft palate, *Am. J. Dis. Child., 106,* 185–188, 1963.

(The) Social Security Act and Related Laws (December 1978 Edition), Committee on Finance, U.S. Senate, December 31, 1978.

Wallace, H. M.; Cohen, R; and Siffert, R., Advisory Committees and Consultants in Programs for Crippled Children, *Public Health Reports, 83(2),* 652, 1968.