Submucous Cleft Palate: Prevalence In A School Population

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Introduction

Since Roux first described submucous cleft palate (SMCP) in 1825 (*11*) there have been numerous reports in the literature discussing the symptomatology and treatment of this defect (*1, 2, 3, 5, 7, 9*). None of these, however, contains any information about the prevalence of SMCP in a general population although it has been estimated to comprise 3 to 5% of a Cleft Palate Clinic population (*2, 5, 9*). Early surgical intervention has been recommended despite the report by Kelly in 1910 that a significant percentage of persons with SMCP are asymptomatic (*7*). It would seem important to know the prevalence of this defect in a random population and the frequency of complications before recommending early surgical correction. For this reason 10,836 Denver school schildren were screened for any palatal abnormality and particularly for SMCP and bifid uvula (BU).

Methods

This project was conducted jointly by the Cleft Palate Team of the Birth Defects Clinic (BDC), University of Colorado Medical Center and by the Division of Health Services, Denver Public Schools. It extended from May 1, 1968 through January 31, 1970 and included 15 school months. Prior to this time, a special teaching session was held for the physicians who would be examining the school children and SMCP was discussed and demonstrated. At this time, they were given the opportunity to examine a child with SMCP to compare their findings.

The school physical examinations were done for a variety of reasons, in most cases "routine," and were done on kindergarten through high school age children. Physician participation in this project was voluntary. During the examinations, the cooperating physicians examined the oral cavity

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both by inspection and by palpation. They were asked to record any suspected abnormality of the lips, palate or speech and to note whether the uvula was bifid or single. The records were then sorted, separating those with any suspected abnormality. The parents of the children with suspected abnormalities were contacted through the schools and permission was obtained to repeat the oral examination. Each child was examined independently by two of the three authors and their findings compared; the degree of correlation between the two examiners was high. At this time a specific diagnosis was made. Where differences of opinion occurred or where a SMCP was suspected, the parents and the family physicians were both contacted and the family asked to come to the BDC for further evaluation and final confirmation of the diagnosis. Details of this evaluation have been reported elsewhere (*12*).

Results

Ten thousand eight hundred thirty-six children were screened; in 237 some abnormality or a bifid uvula was suspected. These "abnormalities" are described in Table 1. Thirty-four children were not examined; either permission for further examination was denied or the child moved, etc. Thirty-seven were normal on re-examination by the SMCP school team. One hundred children had an isolated BU, 8 had a BU in association with SMCP and 2 others had an associated cleft lip. One additional SMCP was detected without a BU for a total of 9 SMCP, none of which had been previously diagnosed. Seventeen children had repaired cleft lip and/or cleft palate or isolated cleft palate. The remaining children had a variety of other abnormalities including dental problems, high arched palates,

TABLE 1.

Total Number Screened			10,836
Normal		10,599	
Suspected Abnormality		237	
Normal		37	
Not Examined		34	
Submucous Cleft Palate		9	
With Bifid Uvula	8		
Without Bifid Uvula	1		
Isolated Bifid Uvula		100	
Primary Speech Problems		13	
Repaired Cleft Lip and/or Cleft Palate		15	
Cleft Lip with Bifid Uvula	2		
Repaired Cleft Palate		2	
Primary Dental Problem		2	
Other Palatal Abnormalities		25	
Treacher-Collins Syndrome	1		
First Arch Anomalies	1		
Total		237	

TABLE 2.

	sex	*ethnic group*	*age*	*bifid uvula*	*audiogram*	*speech*
J.O.	F	C	10	+	N	N
L.S.	M	SA	13	−	N	N
W.M.	M	C	17	+	N	N
L.K.	F	C	18	+	N	N
T.F.	F	SA	8	+	?N	N
B.E.	F	C	11	+	N	N
R.L.	F	C	13	+	N	A
P.K.	M	C	15	+	N	N
D.M.*	F	SA	6	+	?	?

* Refused further evaluation.
C—Caucasian; SA—Spanish surname; N—Normal; A—Articulation errors.

torus palatini, short palates and/or speech defects. One child had Treacher-Collins syndrome, one had multiple first arch anomalies and several had other minor anomalies such as hypertelorism or clinodactyly.

The characteristics of the 9 children with SMCP are summarized in Table 2. There were 6 females and 3 males, 6 Caucasians and 3 with Spanish surnames. One family refused further evaluation (D.M.). No child had a history of recurrent serous otitis media although one girl (T.F.) had bilateral myringotomy tubes in place and a borderline normal audiogram. Speech was normal in 7 while 1 had articulation errors. None had hypernasality. Examination of family members revealed a similarly affected sibling in one case (W.M.).

Discussion

The incidence of cleft lip and/or cleft palate and of isolated cleft palate has recently been reviewed by Fraser (*4*) and the frequency of BU has been studied in various populations (*6, 8, 10*). To the authors' knowledge, however, no similar study has been made of the incidence or prevalence of SMCP. Our project has detected 9 SMCP among 10,836 Denver School children for a prevalence of 1:1,200 in this population.

There are several potential problems in a study of this sort. The most obvious is the number of physicians involved in the screening process and the possibility that some SMCP were missed. Those physicians who participated did so voluntarily and enthusiastically. The recording system was kept very simple and the physicians were asked to indicate only any suspected abnormality or variation and not to make a definitive diagnosis. Although inter-examiner reliability was not checked, we feel that the simplicity of the recording system and the subsequent tendency to "over-diagnosis" led to the notation of most abnormalities or variations from normal. The specific diagnosis was made not by the school physicians but by the authors and the full SMCP team.

In addition, the physicians were asked to indicate the presence of a BU

because of its frequent association with SMCP and as a check on the accuracy of the study. The prevalence of BU was about 1:100 in this study as compared with 1:75 reported by Meskin, et al (*8*). When one considers that 34 children with suspected abnormalities were not examined and that among these would be additional cases of BU, it is apparent that the 1:100 is a minimal figure which compares favorably with the 1:75 previously reported. It is also possible that a SMCP was present but not diagnosed in the group of children that were not examined by the authors. It would be highly unlikely that more than one SMCP would have been missed in this way. The authors were very impressed with the cooperation and enthusiasm of the participating physicians and feel that few if any cases of SMCP were missed by the physicians screening the students.

There is also a problem in adequately defining the population screened. The Denver school population during the years 1968–1970 was 66% Caucasian, 14% Black, 19% Spanish surname and less than 1% Oriental. The prevalence figure of 1:1,200 would, therefore, apply to a mixed population representing a spectrum of socio-economic classes and a variety of ethnic groups. The authors agree that the ideal study would be confined to a single ethnic or racial group but choose the more diverse school population because of its availability. Future studies of single populations should be encouraged.

A school population was also selected because of its inclusiveness. The city of Denver has classes for the educably retarded and for the physically, visually and hearing handicapped. It was, therefore, felt that the group screened would be representative of all but the most severely handicapped. Our personal experience has shown that SMCP may be one of multiple abnormalities in a variety of syndromes and these may have been excluded from the screening process. The primary goal of this study, however, was to determine the prevalence of SMCP in a "normal" population and the school population fulfilled these criteria.

The major difficulty encountered in this study was the actual definition of a SMCP. The three classic features are (1) a bifid uvula, (2) diastasis of the palatal muscles and (3) a palpable notch in the hard palate (*9*). This study has demonstrated that there is a spectrum of abnormalities and that the diagnosis may not be clear-cut, a point emphasized by Crikelair, et al (*2*). We defined a SMCP as a palate with 2 of the features described above and the final diagnosis was made by a team consisting of pediatricians, plastic surgeons, an otolaryngologist, dental sub-specialists and the public health nurse. This definition was reached after reviewing the literature and consulting with those persons who had had previous experience with this defect.

The primary purpose of this study was to obtain some information about the prevalence of SMCP in a random population. Prior to this survey no figures were available. The authors acknowledge some statistical impurities in this study. We feel, however, that the point has been

made: SMCP is not a rare abnormality and in a significant number of persons it is asymptomatic and unrecognized.

Summary

Ten thousand eight hundred thirty-six Denver School children were screened for abnormalities of the oral cavity or speech and 9 cases of SMCP were detected. All were previously undiagnosed and none had significant symptomatology. This is the first report of the prevalence of SMCP in a general population.

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