Longitudinal Speech Performance in Patients with Cleft Palate: Comparisons Based on Secondary Management

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Speech performance of three groups of speakers with cleft palate was analyzed at two-year intervals from age 4 years through age 16 years. The three groups consisted of: (1) Group A—individuals who score below 20 percent correct on the Iowa Pressure Articulation Test (IPAT) at 4 years of age and who received secondary surgery by 8 years of age; (2) Group B—individuals who scored below 20 percent correct on the Iowa Pressure Articulation Test at 4 years of age, but did not receive secondary management by 8 years of age; and (3) Group C—individuals who scored above 20 percent correct on the Iowa Pressure Articulation Test and who did not receive secondary surgery by 8 years of age. The results demonstrated that Group A performed better on several measures of speech performance at 16 years of age than either of the other groups. Also, speech performance appeared to become slightly worse around 12 years of age for Group B. This may be interpreted as evidence that this group may have been negatively affected by the physiologic changes associated with puberty. The other two groups did not appear to show this effect. The results of this study emphasize the need for careful research regarding patients who, in spite of early poor speech performance, are not considered candidates for early secondary management.

Secondary surgical procedures such as pharyngeal flap and Teflon injection are frequently recommended for speakers with cleft palates in order to improve velopharyngeal function for speech. Such procedures are usually performed after 4 years of age and may be delayed until puberty or later if the patient's prospect for nonsurgical improvement is not well understood.

Secondary management is generally believed to improve speech production, but studies have shown that such procedures do not guarantee normal articulation (Riski, 1979; Van Demark, 1979; Van Demark and Hardin, 1985). For example, Van Demark (1979), in a retrospective study, considered three subgroups of cleft palate speakers identified on the basis of speech performance and surgery. Group A consisted of those subjects who scored below 20 percent correct on the Iowa Pressure Articulation Test (IPAT) at 4 years of age and who later were given secondary surgical management for velopharyngeal incompetence. Group B consisted of those subjects who scored below 20 percent correct at 4 years of age on the IPAT but who received no secondary management by age 8 years. Group C consisted of those subjects who scored 20 percent or better on the IPAT at 4 years of age and who received no secondary management. The results of that study indicated that the group who required secondary management had a significantly higher incidence of nasal distortions at 4 years of age compared to the two other groups. Also, Group A had poorer articulation scores at 8 years of age, but by 10 years of age was judged to have better velopharyngeal function than either of the two other groups.

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The purpose of this investigation was to reexamine the Van Demark 1979 subjects through age 16 years in order to determine if speech characteristics remained constant or improved with age in each group. Specifically, we hypothesized the following: (1) Those individuals with poor speech and poor velopharyngeal closure at 4 years of age (Group A) would achieve, after secondary surgery, good speech and good velopharyngeal closure by 16 years of age. (2) Those individuals with poor speech and good velopharyngeal closure at 4 years of age (Group B) would achieve good speech and good velopharyngeal closure by 16 years of age. (3) Those individuals with good speech and good velopharyngeal closure at 4 years of age (Group C) would achieve good speech and good velopharyngeal closure by 16 years of age.

METHODS

The subjects included in the Van Demark (1979) report were reanalyzed in a manner similar to that employed in the original report for subjects at 4, 8, and 10 years of age. However, for the purpose of this project, additional data were included for each group at 6, 12, 14, and 16 years of age. Also, in this study we avoided the previously employed method of including data obtained from a subject "nearest to" each age group. Our more conservative approach resulted in some small but observable differences in group size for some age categories compared to the previous report. Therefore, the average scores for some groups in this report may be expected to differ slightly from the scores provided about the same groups in the Van Demark (1979) study.

Subject Selection

Criteria for subject selection were identical to that described in Van Demark (1979). Briefly, all 106 subjects included were to have had articulation testing performed at 4 and 8 years of age. No subjects had secondary procedures such as pharyngeal flap or prosthetic management prior to 4 years of age.

The subjects were divided into three groups on the basis of their performance on the Iowa Pressure Articulation Test (IPAT) at 4 years of age and subsequent secondary management. Group A (N=31) consisted of all subjects who scored below 20 percent correct on the IPAT at 4 years of age and who subsequently had secondary management by age 8 years. Group B (N=24) consisted of all subjects who scored below 20 percent correct on the IPAT at 4 years of age, but who did not require further palatal surgery as of age 8 years. Group C consisted of all subjects who achieved 20 percent or better on the IPAT at age 4 years and who did not require secondary surgery as of age 8 years. Distribution of cleft type by group is presented in Table 1. As may be expected, Group A included a higher percentage of subjects with more extensive clefts, i.e., complete unilateral and bilateral cleft lip and palate than did Groups B and C. In Group A, 84 percent of the subjects had either unilateral or bilateral clefts of the lip and palate.

Articulation Testing

The 149-element articulation test described by Van Demark and Tharp (1973) was administered to each subject. Standard articulation testing procedures were employed. For the purpose of this research, the results of that testing were considered in terms of percent occurrence of nasal distortions, oral distortions, glottal stops, pharyngeal fricatives, omissions, and substitutions observed for plosives, fricatives, and affricates.

Each group's mean score was examined also on the 43-item Iowa Pressure Articulation Test (IPAT), a subtest of the Templin-Darley Test of Articulation designed to test consonant sounds associated with relatively high oral air pressure (Morris et al, 1961). Those scores are reported in terms of percent correct. The IPAT has been reported to be a good predictor of the need for secondary management (Van Demark et al, 1975; Van Demark and Morris, 1977). For example, Van Demark et al (1975) found that the IPAT was 95 percent correct when used to dis-

TABLE 1 Type of Cleft Distribution Across Groups

		Soft Palate Only		*Hard and Soft Palate Only*		*Unilateral Complete*		*Bilateral Complete*	
Group	*N*	*N*	*%**	*N*	*%*	*N*	*%*	*N*	*%*
Group A	31	0	0	5	16	15	48	11	36
Group B	24	2	8	4	17	11	46	7	29
Group C	51	7	14	14	27	26	51	4	8

* Percentage of subgroup.

criminate between those who required secondary management and those who did not among a group of 75 patients with cleft palate.

Ratings of Velopharyngeal Competence

Composite ratings were collected regarding velopharyngeal competence (Morris and Smith, 1962). These ratings were categorical in nature with three categories defined: velopharyngeal competence, marginal velopharyngeal competence, and velopharyngeal incompetence. Subjects categorized as demonstrating velopharyngeal competence were considered to achieve normal velopharyngeal closure for speech. Similarly, subjects categorized as demonstrating marginal competence and incompetence were considered to have only marginal or no ability, respectively, to achieve velopharyngeal closure for speech. Evaluation of velopharyngeal competence was based on the examiner's overall clinical judgment when all the data available for a given evaluation were considered. Therefore, these ratings were a composite rating based on results of an oral examination, articulation tests, conversational speech, stimulability, manometer ratios, and in some cases, lateral still cephalograms.

Ratings of Articulation Defectiveness and Nasality

In order to obtain general, composite information regarding perceived acceptability of speech production in the subjects included here, ratings of articulation defectiveness and nasality were obtained for conversational speech at the same time as articulation testing was performed. These ratings were made by the examiner using a seven-point equal-appearing interval scale where 1=least defective or least nasal, and 7=most defective or most nasal. Reliability of these ratings has been found to be acceptable (Van Demark, 1964; Hardin et al, 1985). Hardin et al found for 25 samples evaluated that test-retest interjudge agreement was within one scale value on 92 percent of the samples for articulation ratings and on 84 percent of the samples for nasality ratings. Intrajudge agreement for the same 25 samples was within one scale value on 100 percent of the samples for articulation ratings and on 92 percent of the samples for nasality ratings.

Secondary Management

The bases for recommending secondary management were described in the original report. Generally, however, the decision was based on the evaluation of the University of Iowa Cleft Palate team, which consisted of speech pathologists, plastic surgeons, otolaryngologists, psychologists, orthodontists, and prosthodontists. In all but one case, the secondary management recommended and subsequently performed was a pharyngeal flap operation. The exception was a Teflon injection in the posterior pharyngeal wall in one patient. Mean age of secondary management in the secondary management group (Group A) was 6 years, 2 months.

Results

The results of this longitudinal investigation are presented in a descriptive manner. Inferential statistics were not employed in the data analysis because of various idiosyncrasies of the data. Specifically, the data sets were not balanced because of inherent problems with longitudinal research, such as patient attrition and missed observations at some ages. The authors urge the readers to bear in mind these factors while considering the description of the results. Attrition is discussed in more detail below.

Attrition Effects

Changes in group size for each age are presented in Table 2. Although the numbers in each group remain constant through age 8 years for each group, decreases in group size are evident at each subsequent age. Estimates of the potential effects of attrition were calculated for each age that data were lost. These estimates, presented in Table 3, were based on the influence of the missing subjects' last available IPAT data on the group mean IPAT score at the age when those

TABLE 2 Group Size Statistics

	Group Size (N)		
Age (yr)	*Group A*	*Group B*	*Group C*
4	31	24	51
6	31	24	51
8	31	24	51
10	29	22	46
12	29	19	43
14	25	15	40
16	19	16	27

TABLE 3 Estimated Attrition Effects on Group Mean IPAT Scores (Percent Correct)

	Age (yr)			
Group	*10*	*12*	*14*	*16*
Group A Number of dropouts	2	0	4	6
Estimated effect on IPAT mean	−4.5	0	+2.0	−1.4
Group B Number of dropouts	2	3	4	2
Estimated effect on IPAT mean	+0.7	+0.4	+3.8	−0.2
Group C Number of dropouts	5	3	3	13
Estimated effect on IPAT mean	+1.0	−1.0	−1.2	0.0

data were available. For example, the effects of the missing IPAT data for the two missing subjects in Group A at age 10 years was estimated based on the effects of the data from those subjects on the group mean at age 8 years. This was done by calculating the group mean at age 8 years both with and without the two subjects' data and then calculating percentage change as follows:

$$\% \text{ change} = \frac{\text{Group mean excluding missing subjects}}{\text{Group mean including missing subjects}}$$

The estimated effect of the missing data on the group mean at age 10 years was obtained by multiplying the group mean at age 10 years by percent change from age 8 years. Assuming the missing data would have had the same effect on the group mean, the estimated effect of the missing data on the group mean at age 10 years was −4.5. That is, because of missing data, the IPAT mean score for group A reported here may be 4.5 points lower than it might have been had the data for the two subjects actually been available.

Because the assumption of similar effects becomes more tenuous when extended beyond the age at which the data for a given subject was first missing, effects were not carried over to later ages even if the missing subject continued to be missing. Thus, any potential error in these estimates is likely to increase as age progresses.

As shown in Table 3, the estimated attrition effects were relatively small. The estimates range from −4.5 for Group A at age 10 years to 3.8 for Group B at age 14 years. Because Groups A and B were smaller than C, the estimated effects of attrition as calculated here may be expected to be smaller for Group C, even at ages when the absolute number of subjects lost was greater. These estimates may be interpreted as evidence that neither the best nor the worst performers in any group were more likely to drop out than those who performed more closely to the group's central tendency. This interpretation is based on the relatively small changes observed and the dispersion of those changes around the group means. Attrition effects as calculated here were considered rough estimates and were not used to adjust the group mean data presented below.

Late Secondary Management. Five subjects who had not received secondary management by age 8 years did require management at a later age. Four of these subjects were from Group B. These subjects had clefts of either the hard and soft palate or the soft palate only, and all had primary surgical management consisting of the Von Langenbeck procedure. Mean age of secondary management was late (13.4 years) and consisted of pharyngeal flap for three subjects and Teflon injection for one.

One subject from Group C received late secondary management. This subject was 12 years, 10 months of age at the time that he received pharyngeal flap surgery.

In the interest of consistency, these subjects were not excluded from the data from Groups B and C after secondary management. That they received secondary management after age 8 years should be considered part of the results of this investigation. The small number of patients (less than 7 percent) from Groups B and C who required secondary management supports the validity of the original classification criterion.

Articulation Data

Iowa Pressure Articulation Test. Data for each group and age from the IPAT are presented graphically in Figure 1. As expected, at age 4 years Group C demonstrated the highest scores and Group A the lowest. All three groups displayed a dramatic improvement by 8 years of age with the differences between the three groups shrinking considerably. By 10 years of age, Group A's mean score had caught up with Group B's, and by age 16 years, exceeded the other two groups. Mean percent correct at age 16 years on the IPAT were, for Group A, 88.5 percent, for Group B, 77.5 percent, and for Group C, 82.7 percent. However, none of the three groups at

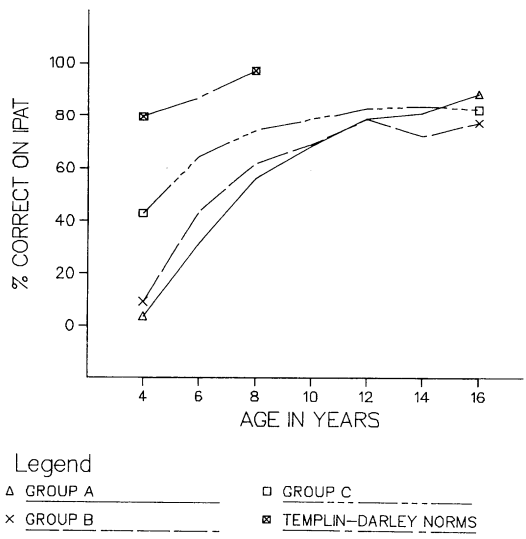


FIGURE 1 Mean percentage correct on the Iowa Pressure Articulation Test for each group and age. Templin-Darley normative data are included at ages 4, 6, and 8 for reference.

age 16 years achieved a mean score equal to the Templin-Darley normative data reported for 8 years of age.

Of possible interest is the slight decline of the articulation score for Group B after age 12 years. Such a decline at this age may be an indication that articulation performance in those subjects may have been affected somewhat negatively by the effects of puberty, such as rapid physical growth and adenoid involution.

Type and Manner of Articulation Errors. Percent occurrence of oral distortions, nasal distortions, glottal stops, pharyngeal fricatives, sound omissions, and sound substitutions as were observed from the 149 element articulation test are presented in Figures 2–7. Errors are reported for plosives, fricatives, and affricates only. For all three groups, oral distortions (Fig. 2) were more common on fricatives and affricates than on plosives. This trend is to be expected owing to dental abnormalities and normal developmental lag in developing lingual consonants compared to labial consonants. Groups B and C demonstrated more oral distortions at age 4 years than did Group A. Incidence of oral distortions peaked at 10 years of age for Group A, 8 years of age for Group B, and 6 years of age for Group C.

Nasal distortions (Fig. 3) were clearly more prevalent for Group A compared to the other two groups through age 10 years. This observation is consistent with the previously reported finding (Van Demark, 1979) that early errors for Group A were more likely to be related to velopharyngeal closure problems than were early errors for Groups B or C. Interestingly, however, Group B demonstrated an increase in the incidence of nasal distortions after age 12 years.

Glottal stops (Fig. 4) and pharyngeal fricatives (Fig. 5), errors that are often considered as compensatory articulation because of velopharyngeal incompetency, were relatively rare in all three groups. Glottal stops were more common in Group A through age 8 years and were primarily observed on production of plosives. Pharyngeal fricatives were also more common in Group A, but these occurred primarily on fricatives and affricates. By age 10 years, glottal stops and pharyngeal fricatives were virtually nonexistent for all three groups.

Errors of sound omissions (Fig. 6) were more prevalent in Groups A and B than in Group C through age 8 years, after which such errors were

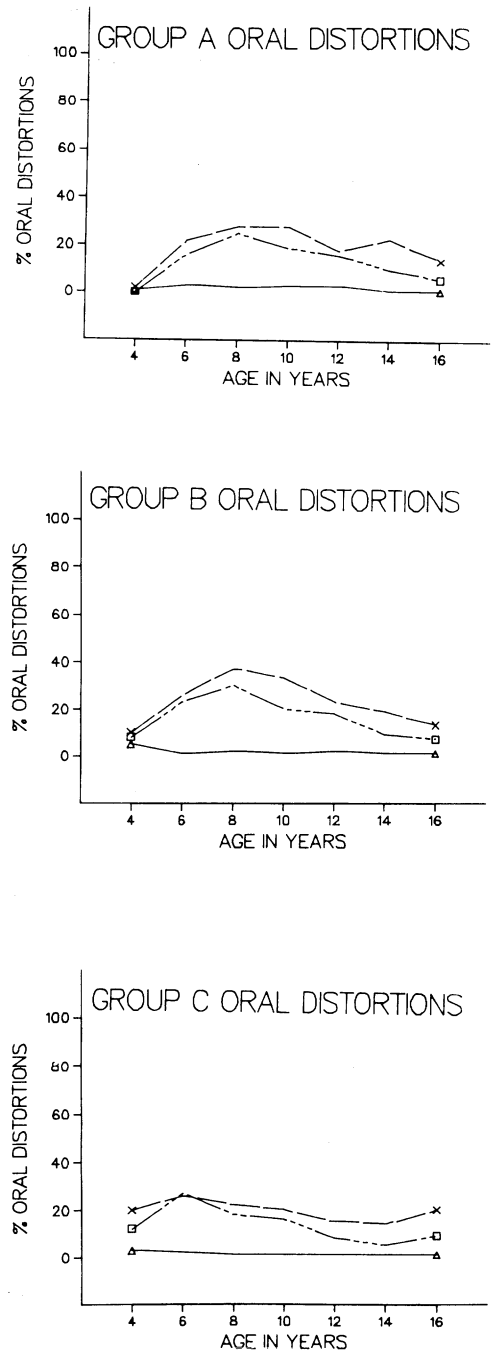
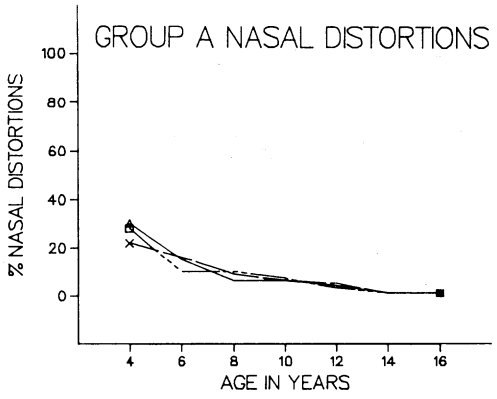
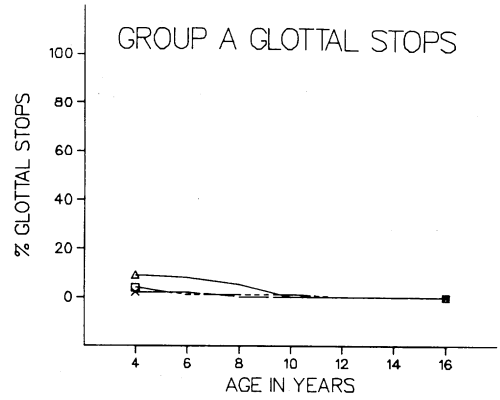
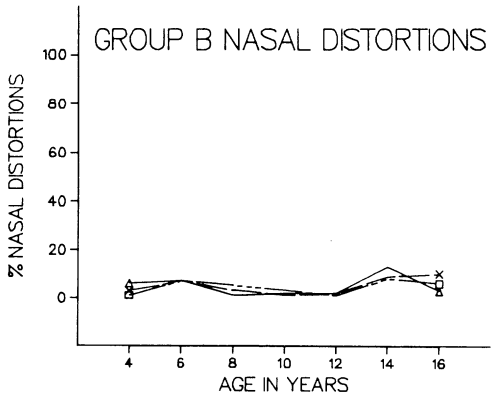


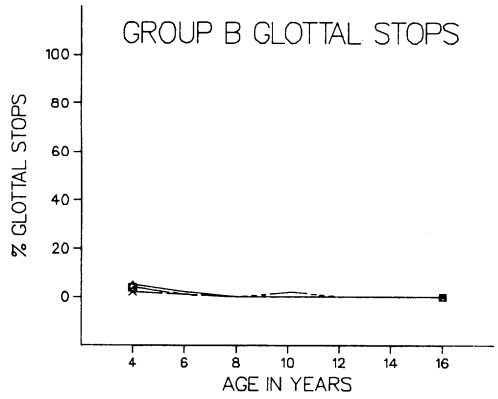
FIGURE 2 Mean percent occurrence of oral distortions on the 149-element articulation test for each group and age.

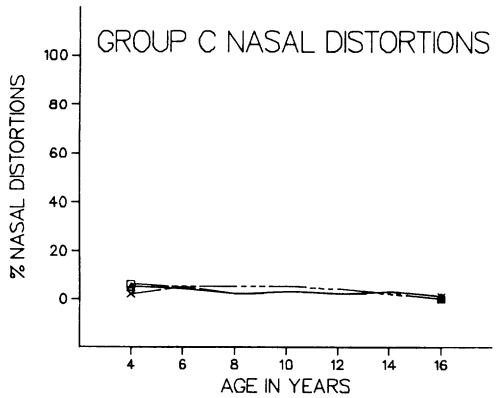
△ = plosives
X = fricatives
□ = affricatives











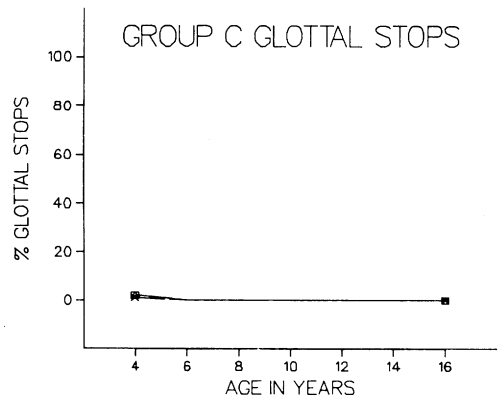
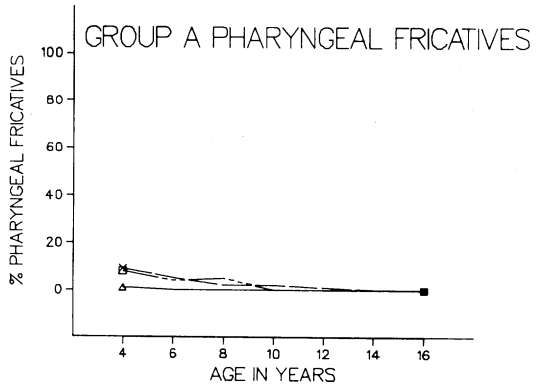


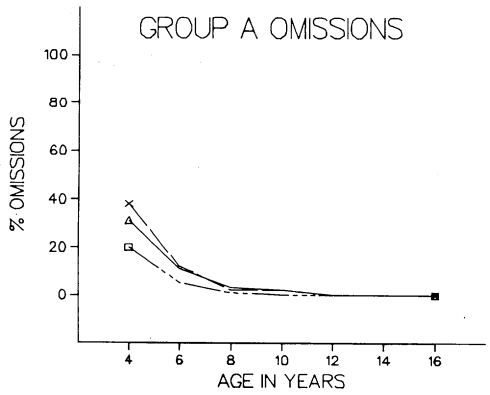
FIGURE 3 Mean percent occurrence of nasal distortions on the 149-element articulation test for each group and age.

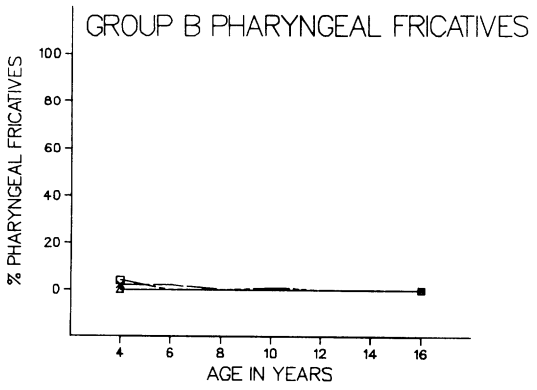
△ = plosives
X = fricatives
□ = affricatives

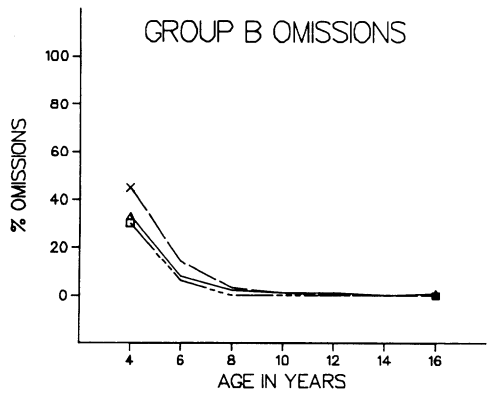
FIGURE 4 Mean percent occurrence of glottal stops on the 149-element articulation test for each group and age.

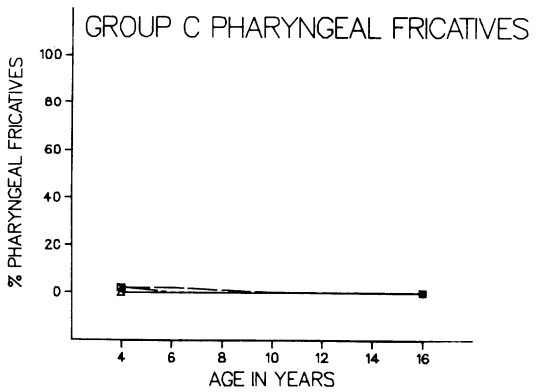
△ = plosives
X = fricatives
□ = affricatives











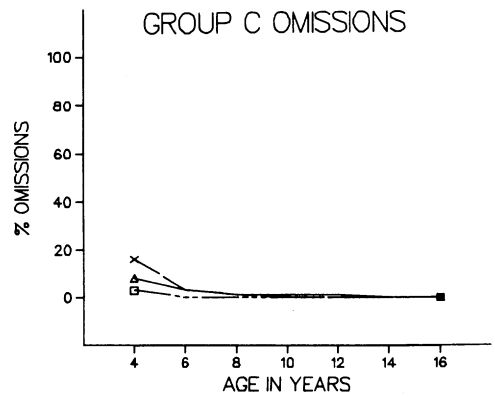


FIGURE 5 Mean percent occurrence of pharyngeal fricatives on the 149-element articulation test for each group and age.

△ = plosives
X = fricatives
□ = affricatives

FIGURE 6 Mean percent occurrence of phoneme omissions on the 149-element articulation test for each group and age.

△ = plosives
X = fricatives
□ = affricatives

rare or nonexistent in all three groups. This is evidence that Group C was able to master many of the double and triple consonant blends (e.g., /sp/, /str/, /spl/) that were included in the test at an earlier age than were Groups A or B. In all three groups, omissions were more common on fricatives followed by plosives and affricates respectively.

Errors of sound substitutions (Fig. 7) were most common in Groups B and C at age 4 years.

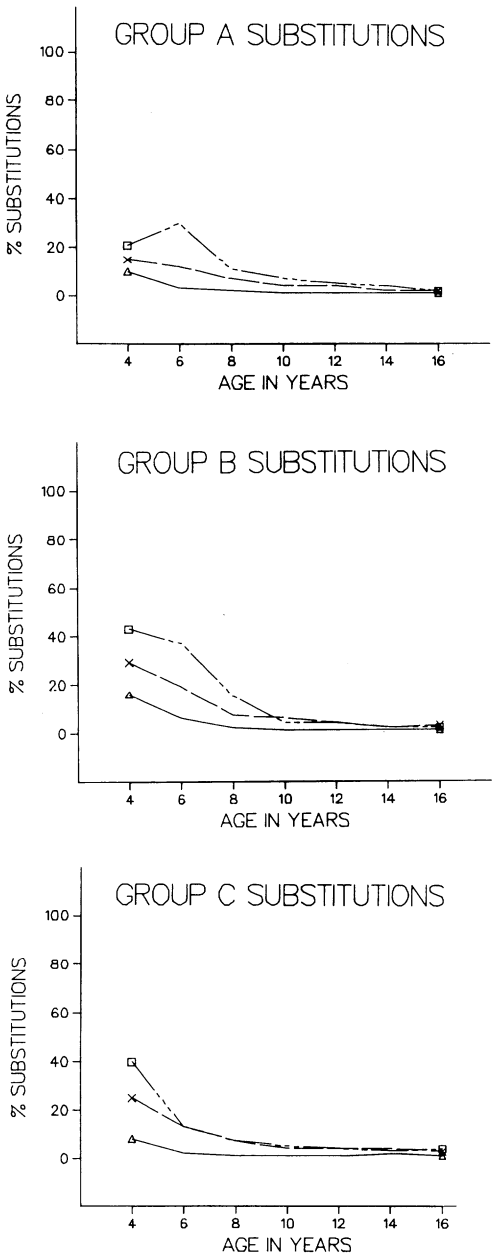


FIGURE 7 Mean percent occurrence of phoneme substitutions on the 149-element articulation test for each group and age.

△ = plosives
X = fricatives
□ = affricatives

These two groups demonstrated approximately twice as many sound substitutions on affricates as did Group A at age 4 years. Group B appeared to be slowest to correct sound substitutions compared to the other two groups in that the incidence of such errors continued to be greater in Group B at age 8 years than in the other two groups. After 10 years of age, substitutions were relatively rare in all three groups.

Ratings of Nasality, Articulation Defectiveness, and Velopharyngeal Competency

Nasality. Mean nasality ratings for each group are presented in Figure 8 (top). The difference between Group A and Group B is most noticeable at age 4 years. The data for Group A showed a marked reduction in mean nasality ratings from age 4 years to age 6 years and again from age 6 years to age 8 years. This is attributed to the effects of secondary management which occurred at these ages. It is of interest that, after age 8 years, nasality ratings for Group A continued to decline systematically to age 16 years. Conversely, after 10 years of age, the mean nasality rating for both Group B and Group C showed an increase beyond Group A followed by a gradual decline. By 16 years of age, however, Group A continued to have a slightly lower mean nasality rating.

Articulation Defectiveness. Ratings of articulation defectiveness, shown in Figure 8 (bottom), indicate only small differences between Group A and Group B at all age groups examined. A small increase for Group B is noticeable, however, at age 12 years. As expected, Group C was rated lower in articulation defectiveness through age 14 years. At 16 years of age, Groups A and C appear to be comparable in articulation defectiveness.

Velopharyngeal Competence. Ratings of velopharyngeal competence for each group are presented in Figure 9. Although, at age 4 years, the number of individuals considered to demonstrate velopharyngeal competence in Group A was zero, by age 16 years a higher percentage of subjects in this group were considered competent than in either Group B or Group C. Group A demonstrated a consistent increase in the number of individuals considered competent across each age examined. Group B and Group C, however, demonstrated an increase of competence but both groups showed a subsequent decline. Group A exceeded the other two groups by age 12 years. Group B had the lowest percentage of individuals considered competent at age 16 years.

Group B's ratings of competence demonstrate the instability of this group over time. For ex-

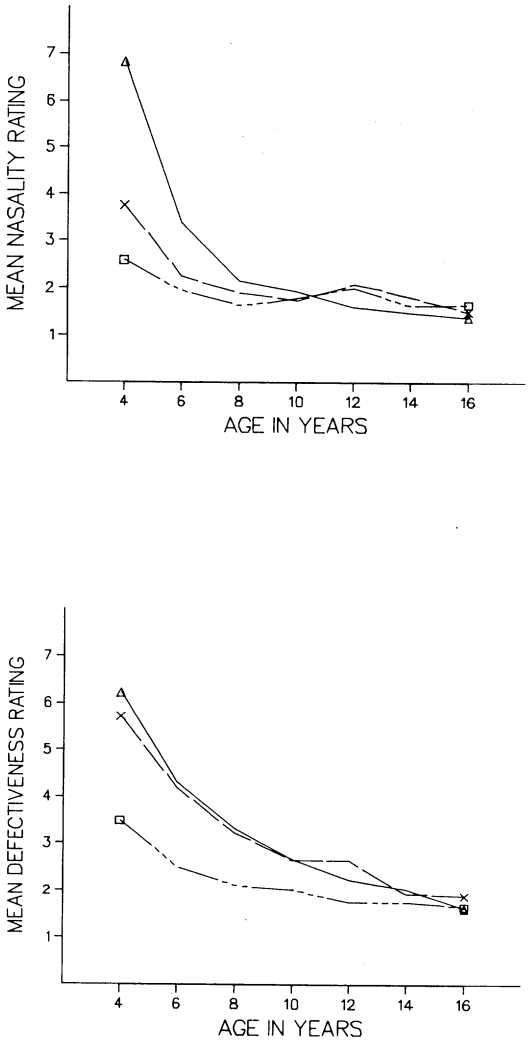


FIGURE 8 Mean nasality ratings (top) and articulation defectiveness ratings (bottom) for each group and age.

△ = **group A**
X = **group B**
□ = **group C**

ample, at age 4 years 37 percent were competent. By age 10 years, Group B's competence increased to 77 percent, followed by a subsequent decline to 56 percent by age 16 years. Conversely, Group B had the highest percentage of individuals considered to demonstrate marginal velopharyngeal competence and also the highest percentage of individuals considered incompetent at age 16 years. As shown in Figure 9 (middle and bottom) the percentage of patients in the marginal and incompetent categories increased after 10 years of age for Group B.

DISCUSSION

Although no single diagnostic measure can be individually considered superior to all others, the results of this study support the strength of articulation performance in discriminating between patients who need secondary management and patients who do not. The use of articulation testing in determining the type of articulation error (i.e., oral distortions and oral substitutions vs. nasal distortions and compensatory articulation) also appears to be diagnostically significant. For example, even though Groups A and B scored below 20 percent correct on the IPAT at age 4 years, Group B errors appeared to be less associated with velopharyngeal incompetence and more associated with learning. No formal test of language development was performed for any of the patients in this study. Our inference with regard to learning is based on the observation that Group B errors at age 4 years consisted largely of errors associated with incorrect learning (omissions and sound substitutions) rather than errors associated with velopharyngeal incompetence (nasal distortions, glottal stops, and pharyngeal fricatives). Future longitudinal studies may benefit from formal measures of language development and learning. The fact that Group B improved without secondary management demonstrates that careful articulation analysis can be beneficial in treatment planning.

These data support several interpretations regarding speech performance in patients with cleft palate and long-term implications of early decisions regarding secondary management. We hypothesized that in spite of the group differences in terms of articulation performance and velopharyngeal competence at age 4 years, each of the groups would achieve similarly good speech by age 16 years. If "good speech" is arbitrarily defined as reaching the norms on the IPAT provided by Templin and Darley (1969), then none of the three groups studied achieved that criterion. Interestingly, the group that had been given secondary surgery by age 8 years (Group A) scored higher on the IPAT at age 16 years (88.7 percent) than either of the other two groups. Group B scored lowest at age 16 years (77.5 percent), and Group C (82.7 percent) fell between Groups A and B.

To our knowledge this is the first time that patients who required secondary management (Group A) have been reported to achieve better articulation and velopharyngeal competence ratings than patients who did not require secondary management (Groups B and C). Although individuals who require secondary management usually have the more severe cleft, this finding demonstrates that once an individual is given an

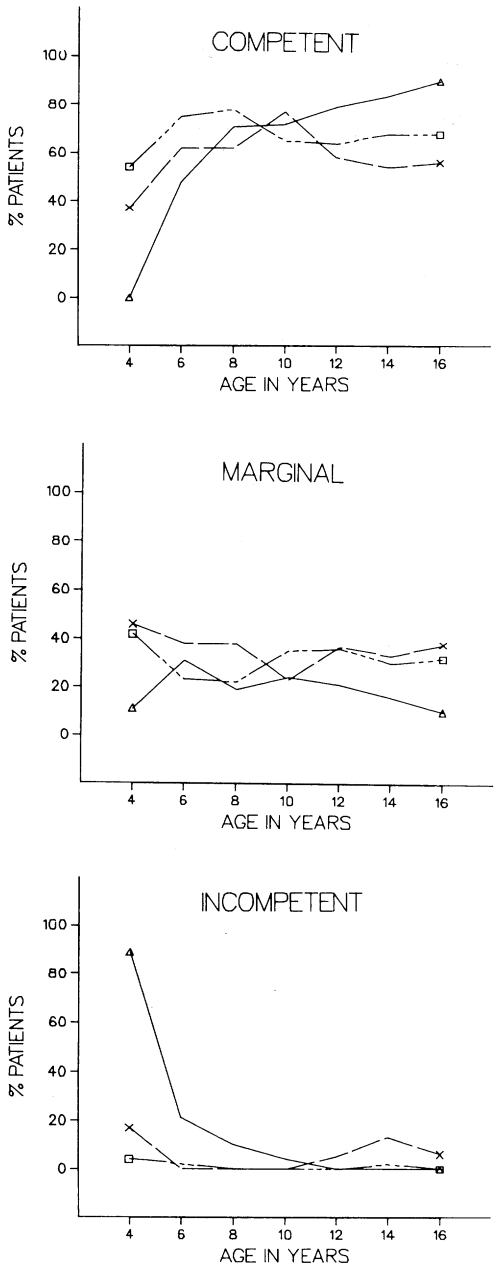


FIGURE 9 Mean percentage of patients rated as competent (top), marginal (middle), and incompetent (bottom) with regard to velopharyngeal closure for each group and age.

△ =group A
X =group B
□ =group C

adequate mechanism, comparable articulation can be achieved.

The three groups appeared to be rank ordered similarly in terms of velopharyngeal competence. That is, the group that received secondary management (Group A), in spite of having no patients considered competent at age 4 years, was found to have the highest percentage of velopharyngeal competence at age 16 years. Group B was found to have the lowest percentage among the three groups of patients considered competent and the highest incidence of patients considered marginal or incompetent at age 16 years. Even Group C, the group with the best articulation and velopharyngeal function at age 4 years, appeared substantially different in terms of velopharyngeal competence at age 16 years.

Group B, in our opinion, probably represents the most difficult management population and should be followed closely. For example, although originally this group presented errors associated with learning or dentition, as age increased velopharyngeal competence appeared to decrease, particularly around the onset of puberty. In fact, four of these patients (16 percent) required late secondary management. Adenoid atrophy, rapid physical growth, and perhaps voice changes associated with the onset of puberty may place the patient with marginal velopharyngeal competence at risk for a decline in speech proficiency. Therefore, subjects with marginal velopharyngeal incompetence should be followed carefully until at least 16 years of age.)

The data show that none of the three groups in this study achieved, on the average, what speech pathologists may consider perfectly normal speech at age 16 years. However, such deficits probably are not so severe by this age as to require intervention. As is shown in Figure 8, mean nasality and articulation defectiveness ratings were approximately two on a seven-point scale. A rating of two may not represent a clinically significant disorder. Further clarification of this issue may be possible by learning more about how the general public perceives speech characteristics indicative of marginal velopharyngeal incompetence. Although such characteristics would necessarily result in error detection by a well-trained speech pathologist, such errors may be acceptable within the speaker's communicative environment. Data such as those presented in this report should be compared not only against established normative data, but also against the speaker's environmental standard.

A recent report (Hardin et al, 1986) described a preliminary attempt to examine the predictability of speech performance at age 14 years in 50 cleft palate children. Although no consistent pattern of speech variables emerged as predictors, gender was found to account for a considerable portion of the variability of the data. The purpose of the current study differed fundamentally from the Hardin et al research in that our purpose was to describe longitudinal speech characteristics. We did not intend to attempt to predict speech outcome statistically. This distinc-

tion between predictive and descriptive purposes is important and makes generalization to this study from the Hardin et al study hazardous (Morris, 1986). We have not considered gender in this research, although future efforts should do so.

Another focus of attention for future research involves those few patients who, at age 15 or 16 years, continue to exhibit velopharyngeal incompetence. Although the numbers reported here are small (at age 14 years, two patients in Group B and one patient in Group C), those patients represent perhaps the greatest challenge to the cleft palate team.

In conclusion, results of this study demonstrated in part that articulation performance for all three groups, as measured with the Iowa Pressure Articulation Test, continued to improve until age 12 years. Differences between the groups were apparent, however, between 12 and 16 years of age. Group A showed steady improvement from 12 to 16 years of age. Group B, during the same time period, showed a decline in performance. Articulation performance for Group C appeared to plateau after 12 years of age. These findings may serve to clarify the findings reported by Van Demark et al (1979). In that report, a group of 351 patients with cleft palate were examined through age 16 years. Although, secondary management was not an issue, the findings from that study indicated that a dip in performance, particularly on fricatives and affricates, was evident at age 13 years. The authors noted, however, that articulation performance continued to improve after 10 years of age. Based on the results of the current study, prospects for continued improvement beyond age 10 years may be dependent on early articulatory performance and history of secondary management.

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