Heterogeneity of the 'Cleft Palate Population' and Research Designs

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'Cleft palate' is widely used as a term of convenience to describe populations of individuals who were born with cleft lips and palates, only cleft palates, and, in some instances, only cleft lips. It has been used to describe populations that include individuals with physically managed clefts as well as individuals with unmanaged clefts. It has been used to describe populations of individuals of all ages and of both sexes coming from a variety of cultural backgrounds and endowed with a wide range of intellectual abilities. Frequently it has been used as an adjective as in the phrase, 'cleft palate speech', and in this context has been used in some instances to describe the speech of a person who has never had a cleft of any kind.

When a single term such as 'cleft palate' is used to refer to a large group of individuals, it is often assumed that these individuals possess homogeneous characteristics. Yet it is obvious that wide individual variations exist within such a group. It is the thesis of this paper that the heterogeneity of the cleft population is too often overlooked with consequences that, in some instances, lead to faulty research designs and misleading research findings. It is the purpose of this paper to explore some of the implications of this thesis.

Cleft Type

First, let us consider the variable of cleft type. Congenital clefts may vary from a bifid uvula or notched lip at one extreme to a bilateral cleft lip and palate or the 'horseshoe-shaped' palate-only cleft with a significant deficiency of tissue. We classify these individuals into such categories as cleft lip only, cleft lip and palate, cleft of the hard and soft palate only, and cleft of the soft palate only. Although it is recognized that such a classification system still ignores variations within the subgroups, it repre-

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sents an improvement over the procedure of combining all cleft types into one group. It is obvious that differences in cleft type exist; however, what is not always recognized is that differences between these subgroups are not restricted simply to the extent of the cleft. There is a growing body of knowledge which demonstrates that these groups are different on a number of parameters.

In the first place, there appear to be differences in articulation skills between the cleft-type groups. Bzoch (*2*), Starr (*14*), Counihan (*3*), and Spriestersbach, Moll, and Morris (*11*) have reported that the palate-only subgroup exhibits a lower mean score on articulation tests than does the lip and palate subgroup. Although some of the reported differences were not found to be statistically significant, and although Byrne, Shelton, and Diedrich (*1*) found differences in the opposite direction, the consistency of the findings from study to study appear to justify the conclusion that under current philosophies of management, individuals with lip and palate clefts, as a group, demonstrate a higher level of articulation skills than do individuals with only palate clefts. In addition, research evidence (*11*) indicates that individuals with only cleft lips have essentially normal articulation skills.

Another systematic difference between cleft-type groups, which is closely related to articulation, involves the ability to impound intra-oral breath pressure. Spriestersbach and Powers (*12*), studying individuals with surgically repaired clefts, found that those with palate-only clefts have lower wet spirometer ratios than those with clefts of both the lip and palate. This finding suggests that the results of physical management, in terms of the adequacy of velopharyngeal function, are poorer for the palate-only group than for the lip and palate group.

Differences in the incidence of hearing loss and ear pathology have also been reported. Masters, Bingham, and Robinson (*6*), and Spriestersbach and others (*10*) found that individuals with clefts of the palate-only have a higher incidence of hearing loss than do those with clefts of the lip and palate. Graham (*5*) reports similar intergroup differences in hearing loss and history of ear disease.

In relation to intellectual functioning, Goodstein (*4*) found that the palate-only group had significantly lower scores on intelligence tests than did the lip and palate group. Although Goodstein did not test the significance of the difference between the lip-only and palate-only groups, inspection of the data reveals that a similar difference in intelligence probably exists between these two subgroups.

Spriestersbach, Spriestersbach, and Moll (*13*) report that 16% of the children with clefts of the lip, and lip and palate, in their study had associated congenital anomalies while 51% of those with clefts of the palate-only had such other anomalies. Rank and Thomson (*8*) report similar differences in the incidence of associated anomalies between these subgroups.

It is apparent, then, that there are a number of systematic differences among various cleft-type subgroups within the cleft population. These differences, and possible others not yet identified, make it imperative that the subgroup or subgroups of cleft types be specified when studying individuals with clefts. For example, when studying the incidence of speech problems in the cleft population, a very real bias is introduced if subjects with only cleft lips are included in the sample. Since the speech of these individuals is essentially normal, the inclusion of them in the subject group, just because they have clefts of some kind, is equivalent to including an equal number of normal, noncleft speakers.

When evaluating physical management results in terms of speech adequacy or speech potential, it is important to consider cleft-type subgroups separately, since it appears that generally better results, in terms of velopharyngeal competence, are obtained for one subgroup than for another. Combination of the groups may tend to obscure important relationships.

When studying the psycho-social characteristics of individuals with clefts, cleft-type groups again should be studied separately. The groups differ on a number of variables, such as cosmetic appearance, intelligence, and speech, which are probably related to psychological adjustment and social skills.

When studying facial growth of individuals with clefts, it also may be necessary to consider type of cleft. It is quite possible that the facial growth characteristics of individuals with clefts of the palate-only may be quite different than those of individuals with clefts of both lip and palate. Differences between these two subject groups on such factors as the degree of alveolar deformity, the difficulty encountered in surgical closure of the palatal cleft, and the presence of other congenital anomalies, feasibly could be related to growth of the facial complex.

Thus, heterogeneity in cleft type may have an effect on various types of research findings and, as a result, should be taken into consideration in subject selection and data interpretation.

Age

Another parameter on which the cleft palate population is heterogeneous is that of age. Studies of this population have included subjects in a wide age range. What may not always be obvious is that many variables which we often study also vary with age. Measures of speech and language status, for example, are dependent, to a large degree, on the age of the subject at the time of evaluation. The data of Morris (7) on 102 cleft subjects indicate that mean length of response, a measure of verbal output, increases systematically with age level. In the same study articulation test scores ranged from 33 at age three and a half to 143 at age 11. Measures of overall speech proficiency, intelligibility, and other verbal communication skills show similar relationships with age.

The importance of these age-related differences in research cannot be ignored. For example, if an absolute measure of speech proficiency on an articulation test is used as the criterion in the measurement of success of physical management for a group of children at a variety of ages, the results may be misleading. Even if management was successful for all subjects, the level of achievement, in terms of articulation skills, would vary with age. In the situation where speech assessments are made before and after the physical management in order to evaluate its success, age is an even more important factor to consider. If too long a period separates the two evaluations, improved speech may be observed even if the management procedure has completely failed; speech improvement may occur solely because the child has grown older in the interim.

Besides speech skills, it also appears that the incidence of hearing loss in the cleft palate population varies with age. Spriestersbach and others (*10*) report greater mean hearing thresholds for children between the ages of 33 and 71 months than for those who were six years or older. Graham (*5*), who had audiograms on the same subjects at two ages, found that the incidence of hearing loss was greater when the children were four to six years old than when they were eight or older.

Many investigators have described the incidence of hearing loss in the cleft palate population and have speculated on the etiological bases of these losses. In general, the findings of these studies are not in very close agreement. A portion of this disagreement may stem from the lack of a single criterion by which hearing loss is defined; however, differences in the age groups studied by different investigators may also be a factor. The procedure of combining age levels in an investigation of hearing loss or ear pathology in this population probably results in a loss of information and in misleading conclusions.

Speech

Another parameter on which individuals with clefts are heterogeneous is that of speech. Almost all investigations indicate that speech may range from normal to completely unintelligible in this population. Again, although this variability is obvious, its implications for research design are often ignored. For example, a number of studies have been carried out in an attempt to identify the acoustic characteristics of nasal voice quality. In a few investigations this has been attempted by analyzing acoustic differences between the speech of cleft palate and normal subjects. Rarely, however, is the precaution taken to insure that the cleft palate subjects actually exhibit nasal quality or that the normal subjects do not. It is tacitly assumed by many that if an individual has a cleft palate, he exhibits 'cleft palate speech', which, as everyone knows, is characterized by nasal voice quality. Yet, it is obvious that not all individuals with clefts are nasal and, conversely, that not all noncleft individuals have normal voice quality. As a result, investigations of the acoustic characteristics

of nasality in which this fact is not recognized are not efficiently designed and may lead to erroneous results.

The wide range of speech characteristics in the cleft population is also a consideration in studies designed to test the hypothesis that some of the speech problems of individuals with clefts are related to abnormal tongue carriage or tongue movements. In this instance, it is important that the subjects studied exhibit speech problems in addition to having been born with a cleft palate. It might also be pointed out that individual variations in dentition, arch development, velopharyngeal adequacy, and other variables which could be related to tongue carriage and movement also should be considered in such a study. Yet, in most investigations of lingual abnormalities, such variables have not been taken into account. In our opinion, the controversial evidence that exists concerning tongue carriage and function in individuals with clefts is not so much the result of inadequate instrumentation or unreliable observations but rather of the fact that experimenters have not given due consideration to the need for homogeneity among their subjects.

A Specific Illustration

We have seen that the cleft palate population is quite heterogeneous in relation to cleft type, age, and speech proficiency and that these variations are often related to the variables being investigated. There are, of course, many other parameters, such as type of physical management, intelligence, and socioeconomic status, on which wide variability within the cleft population can be demonstrated. It is not the purpose of this paper simply to demonstrate that such heterogeneity exists; in most instances it is obvious. However, it should be pointed out that even the obvious often is ignored. For example, let us consider a specific study, that of Spriestersbach, Darley, and Rouse (*9*). These investigators studied the speech sound articulation of a group of 25 subjects who were heterogeneous on a number of factors. The closure of the palatal clefts of eight of the subjects had not been completed at the time of the study, and one subject had a cleft of the lip only. Five of the subjects were rated as having dentition which the judges were certain would have an adverse effect on speech and 12 were rated as having velopharyngeal mechanisms which would have an adverse effect on speech. Some of the subjects were clearly of an age where a number of the articulation errors could be expected to occur because of the normal processes of learning rather than because of the presence of a cleft. Some of the subjects were at a stage of habilitation where the status of the articulatory structures was within the normal range. It is clear that the most that can be said about this population is that each of the subjects had had a cleft of the lip or palate.

It is legitimate to ask for what purpose shall we accumulate data such as those reported by Spriestersbach, Darley, and Rouse? Are we interested in describing the status of speakers with palatal clefts prior to our efforts

to provide anatomical and physiological adequacy? Do we want to study the reasons for the articulation errors that we observe in these patients at various stages of physical habilitation? Do we wish to use the information to plan a program of speech training? These are but a few of the reasons why we might wish to have information about the patterns of articulation in the speech of patients with clefts. The purposes of the study of Spriestersbach, Darley, and Rouse are not clear. But the purpose for our observations must be considered for it will determine the specifications which we place on our sample and on our analyses of the data. Such specifications, appropriately and thoughtfully drawn, will insure that we will obtain data that can be used with directness and certainty to answer the questions implicit in our purpose.

Summary

The primary point made in this paper is that we must reject the notion that there is some inherent, universal commonality among individuals who are born with a cleft lip and/or cleft palate. We must recognize the various pertinent subgroups within this population. This does not mean, however, that one must always recognize the smallest subgroup in doing a given study. On the contrary, we would defend the use of fewest possible subgroups consistent with the specific research purpose. Nevertheless, we feel that time spent considering the purposes of a study and the subgroups which need to be identified will tend to minimize the number of contradictory results, and will assist us in the interpretation of the observations that our insight and technology make possible.

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References

1. Byrne, Margaret C., Shelton, R. L., Jr., and Diedrich, W. M., Articulatory skill, physical management, and classification of children with cleft palates. *J. speech hearing Dis., 26,* 326–333, 1961.
2. Bzoch, K. R., An investigation of the speech of pre-school cleft palate children. Ph.D. dissertation, Northwestern Univ., 1956.
3. Counihan, D. R., A clinical study of the speech efficiency and structural adequacy of operated adolescent and adult cleft palate persons. Ph.D. dissertation, Northwestern Univ., 1956.
4. Goodstein, L. D., Intellectual impairment in children with cleft palates. *J. speech hearing Res., 4,* 287–294, 1961.
5. Graham, M. D., A longitudinal study of ear disease and hearing loss in patients with cleft lips and palates. M.S. thesis, Univ. of Iowa, 1962.
6. Masters, F. W., Bingham, H. G., and Robinson, D. W., The prevention and treatment of hearing loss in the cleft palate child. *Plastic reconstr. Surg., 25,* 503–509, 1960.
7. Morris, H. L., Communication skills of children with cleft lips and palates. *J. speech hearing Res., 5,* 79–90, 1962.
8. Rank, B. K., and Thomson, J. A., Cleft lip and palate in Tasmania. *Med. J. of Australia, 47* (2), 681–689, 1960.

9. Spriestersbach, D. C., Darley, F. L., and Rouse, Verna, Articulation of a group of children with cleft lips and palates. *J. speech hearing Dis., 21,* 436–445, 1956.
10. Spriestersbach, D. C., Lierle, D. M., Moll, K. L., and Prather, W. F., Hearing loss in children with cleft palates. *Plastic reconstr. Surg., 30,* 336–347, 1962.
11. Spriestersbach, D. C., Moll, K. L., and Morris, H. L., Subject classification and articulation of speakers with cleft palates. *J. speech hearing Res., 6,* 362–371, 1961.
12. Spriestersbach, D. C., and Powers, G. R., Articulation skills, velopharyngeal closure, and oral breath pressure of children with cleft palates. *J. speech hearing Res., 2,* 318–325, 1959.
13. Spriestersbach, D. C., Spriestersbach, Bette R., and Moll, K. L., Incidence of clefts of the lip and palate in families with children with clefts and families with children without clefts. *Plastic reconstr. Surg., 29,* 392–401, 1962.
14. Starr, C. D., A study of some of the characteristics of the speech and speech mechanisms of cleft palate children. Ph.D. dissertation, Northwestern Univ., 1956.