SUMMARY

A study was undertaken to investigate the applicability of utility theory to cleft lip and palate treatment decision making. Questionnaires were used to assess value judgments and attitudes toward risk from 119 individuals associated with seventeen cleft lip and palate treatment centers. Significant differences were found to exist between the values expressed by clinicians grouped according to their specialty or the facility to which they were affiliated. Fathers of cleft lip and palate children differed from cleft palate teams in their judgments regarding the relative values of speech and cosmetic treatment outcomes. No such differences were found between team members and mothers, however. It is suggested that these differences affect the desirability of treatment decisions and the outcomes that follow from them. Therefore, cleft lip and palate treatment planning and the evaluation of alternative therapeutic interventions might be based on a mathematical theory of decision making that explicitly incorporates the subjective assessment of a child's habilitative needs and the goals of the habilitative process.

The Mathematics of Cleft Lip and Palate Treatment Evaluation: Measuring The Desirability of Treatment Outcomes

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Introduction

The child with a cleft lip and cleft palate faces an uncertain future. The trauma of repeated surgery, the stigma of being "different," and the drama of long term-care are the parameters that may well shape future years. Yet, despite cosmetic, dental, speech, hearing, and psychosocial handicaps, the habilitative potential is unequalled by many other defects that are as severe. Rarely are there defects so handicapping to the child, or so disturbing to the family, and yet so amenable to treatment (Pruzansky, 1953). However, not all cases reach the high level of success that modern treatment often makes possible. The fact remains that there are treatment failures. Deciding the success or failure of a treatment or a series of treatments is not a simple matter, however. A 1973 state-of-the-art review of clinical research in cleft lip and palate concluded that there do not exist

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adequate techniques to document, evaluate, and establish the degree of handicap either before treatment or after treatment (Spriestersbach, et al, 1973).

This poor ability to document outcomes is, perhaps, reflected in the organization and structure of cleft lip and palate services. More than 34 disciplines and subspecialties have been catalogued in connection with cleft lip and palate treatment. Since the services of one specialty are frequently determined by the habilitative program being provided by another specialty, interdisciplinary approaches to treatment planning and decision making have become popular.

One finds relatively little variation in the types of disciplines represented on cleft palate teams. However, they do differ in their approaches to and timing of therapeutic intervention. The literature reflects the variety of treatments that are being used by teams, but it generally fails to resolve the uncertainty and controversy that surrounds the selection of the most effective treatment program. Some variations in treatment programs can be explained by differences in the availability of certain skills or the proficiency a particular operator might have with a certain technique. Other variations may have their roots in the inability to document outcomes for treatment evaluation.

This study attempts to describe an approach to the formulation of techniques by which to document a treatment outcome and to relate that outcome to the degree of handicap so that treatment evaluation might be possible. The methods and results described herein represent the application of a mathematical theory of utility first put forth by Von Neumann and Morgenstern (1947). The theory details a formal approach to the assessment of values which can be assigned to treatment outcomes that permit the evaluation of decision alternatives. These values are specially determined to reflect a decision-maker's own views as to the degree of handicap that is present. Also, the values incorporate the decision-maker's attitude towards the element of risk or chance that must be faced with any treatment decision. Since the values are a function of a particular clinician's preferences for certain treatment outcomes and of his own concept of acceptable risks and of justifiable clinical investment, they are necessarily relative. Indeed, the approach presented here takes cognizance of the fact that there are no entirely objective indices by which to measure the habilitative success of treatment in terms of the residual degree of handicap that is present. Rather, one must look towards relative values, on an individual basis, that determine the acceptability of both a treatment decision and its outcome. Koepp-Baker (1971) suggested such an approach when he wrote:

> There are often situations in which the acceptability of the decision, and the actions which follow from it, may not be great or may not exist at all. Families do refuse to accept certain surgical procedures, neglect or refuse to go to a dentist, or find practical excuses why speech therapy cannot be provided. A large number of situational factors—cultural, religious, levels of sophistication, education—may frustrate the best efforts at therapeutic programming though they may be of both high quality and acceptance as judged by the team.

> It is necessary, therefore, that some member of the team (often the medical-social worker) represent these interests and facts in the decision-making process and also in program execution.

Of course, one must recognize that team members are not immune to the same set of factors that determine the family's preferences and which are reflected in their acceptance of a decision. Each specialist on the team may have a different viewpoint regarding the habilitative needs of the patient. Attitudes that describe acceptable levels of risk and treatment outcomes may also vary. Precisely whose attitudes and preferences it is that frustrates the "best" efforts at therapeutic programming, is an open question.

If differences in relative values which govern the acceptability of certain decisions and the outcomes that follow from them can be shown to exist, then they can add considerably to our understanding of variations in treatment programs. Most importantly, if the theory of utility can be shown to be reasonable for cleft lip and palate, then it can begin to form the basis of a mathematical approach to treatment evaluation. It should be noted that utility theory, a component of a field known as decision analysis has been successfully applied to the evaluation of decision alternatives in business (Swalm, 1966) in government (Keeney, 1973) and in other fields of medicine (Betaque, 1969; Forst, 1970; Giauque, 1972; Ginsberg, 1971; Pliskin, 1974). However, the results given here represent the first attempt at applying utility theory to cleft lip and cleft palate.

Methods

Utility theory deals with decision-making under uncertainty; that is, even though a decision-maker may choose a treatment alternative, some element of chance or risk enters into the actual outcome which is to follow from that decision. Utility theory provides a technique by which to associate specially constructed values (called utilities) with treatment outcomes. It is possible, therefore, to construct a scale over the outcomes that evaluates them according to the decision-maker's preferences. Further, if one is consistent with the axioms upon which the theory is based, then a decision-maker's preferences in situations with risky outcomes are completely described by the expected value of the utilities of that situation's possible outcomes. Hence, utility theory is a normative theory of decision making and contains within it the ability to evaluate alternative treatments even though the outcome of a treatment decision can be described only as a range of possible results.

As an example, consider speech as one aspect of a cleft lip and palate treatment outcome. For simplicity, speech is measured according to its intelligibility, the percentage of spoken words, in a passage of connected speech, accurately identified by a normal listener (Subtelny, et al, 1972). Now, consider as a possible therapeutic intervention in a particular case some treatment that has a 50 per cent chance of resulting in speech that is 95 per cent intelligible and a 50 per cent chance of yielding speech that is only 45 per cent word intelligible. Another treatment, in the same case, might have equal chances at 75 per cent

and 65 per cent word intelligibility. Yet another treatment alternative might always result in intelligibility at the 70 per cent level.

If one were to choose among these treatments, assuming that everything about them is the same except their uncertain outcomes, then one would first notice that the *expected* outcome of all of them is the same:

Treatment A: (.5) (95%) + (.5) (45%) = 70% word intelligibility
Treatment B: (.5) (75%) + (.5) (65%) = 70% word intelligibility
Treatment C: = 70% word intelligibility

However, not everyone would agree that these three treatments are equivalent, or, more precisely, not everyone would be indifferent to them. However, by associating a utility value with each level of word intelligibility, it is possible to arrive at a numerical ranking to evaluate the three treatment alternatives consistent with one's own preferences.

Let a utility value of 1 be assigned to the best measure of speech and a utility value of 0 be assigned to the poorest speech. Then:

$$U\ (100\%\ \text{intelligibility}) = 1 \text{ and } U\ (0\%\ \text{intelligibility}) = 0.$$

If there existed a treatment that resulted in either of these two outcomes with equal probability, then the expected utility of such a treatment is calculated to be:

$$(.5)\ U\ (100\%\ \text{intelligibility}) + (.5)\ U\ (0\%\ \text{intelligibility})$$

or:

$$(.5)\ (1) + (.5)\ (0)$$

which is equal to .5. Suppose that a decision-maker has no preference for this treatment or an alternative that always results in 40 per cent intelligible speech. Then the utility associated with 40 per cent intelligible speech is equal to the expected utility of the other treatment, or

$$U(40\%\ \text{intelligibility}) = (.5)\ U(100\%\ \text{intelligibility}) + (.5)\ U(0\%\ \text{intelligibility}) = .5$$

Hence, a utility value of .5 can be associated with 40 per cent intelligible speech. This procedure can be repeated for different values of intelligibility and will generate a number of intelligibility-utility pairs that describe not only the decision-maker's preferences over possible treatment outcomes, but also the decision-maker's attitude toward risk in considering alternative treatments for speech.

Figure 1 illustrates three different intelligibility-utility rankings. The preference ordering is the same for all three hypothetical people; that is, 100 per cent word intelligibility is preferred to 75 per cent word intelligibility, and 75 per cent word intelligibility is preferred to 50 per cent word intelligibility for all of them. However, the three scales describe very different decision-making behaviors. The expected utility values for treatments A, B, and C, considered above, are contained in Table 1.

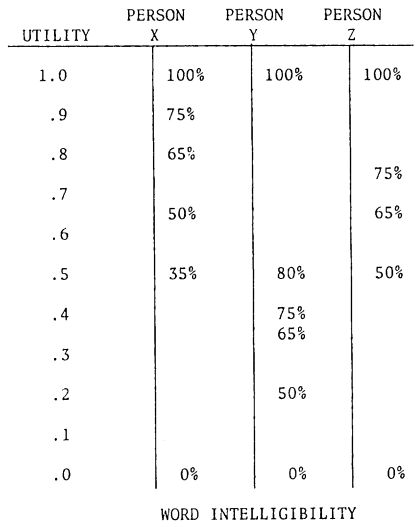

FIGURE 1. Utility functions as indices of desirability.

TABLE 1. Expected untility values for treatments A, B and C, and persons X, Y and Z.

treatment	*person*		
	X	*Y*	*Z*
A	.8	.525	.7
B	.85	.385	.7
C	.9	.375	.7

Inspection of that table reveals that, to be consistent with his own set of values, Person X prefers Treatment C over Treatment B and Treatment B over Treatment A (i.e., .9 > .85 > .8).* But, Person Y's preferences are just the reverse, and Person Z is indifferent. Hence, the utilities are a guide to treatment evaluation since Person X would choose Treatment C, Person Y would choose Treatment A as the "best" or preferred alternative, and Person Z would rank all three treatment alternatives as being equal.

Person Z's behavior is said to be risk neutral since his preferences are represented by the expected value of the outcomes. So, for him, treatments A, B, and C all have the same expected outcome; and he, therefore, is indifferent. Person X, however, exhibits behavior that is said to be risk averse. His preferences increase when the amount of risk decreases. Person Y's behavior is said to be risk-seeking since he places a higher value on situations involving more risk.

Another way to see how these differences in value judgments affect decision making behavior and, ultimately, the evaluation of decision alternatives, is to consider a hypothetical treatment that has a fifty-fifty chance at 100 per cent

* As an example of the calculations that went into Table 1, consider Treatment B for Person X. Person X's utility for Treatment B's outcomes of 75 percent word intelligibility and 65 percent word intelligibility are .9 and .8, respectively. Since Treatment B has equal chances at either outcome, the expected utility for Person X is (.5) (.9) + (.5) (.8) = .85.

word intelligibility or 0 per cent word intelligibility. One would be hard pressed to find such an all-or-nothing treatment, but it serves as a good example by which to discuss the different types of value judgments and decision making behavior that are described in Figure 1. As previously discussed, the expected utility of this treatment is .5. For Person X, 35 per cent word intelligibility has a utility of .5. Hence, Person X, being risk averse, would be indifferent between a treatment guaranteeing an outcome of 35 per cent word intelligibility and the hypothetical example. This is so even though 35 per cent word intelligibility is *less than* the expected outcome of the hypothetical example which is 50 per cent word intelligibility. Person X feels that, for intelligibility above 35 per cent, he would prefer not to choose the hypothetical treatment example and that, for intelligibility of less than 35 per cent he would. This is not true of Person Y, since for him 80 per cent intelligibility has a utility of .5. Person Y, being risk seeking, would prefer the hypothetical treatment example over any treatment that offered less than 80 per cent intelligibility even though 80 per cent intelligibility is *greater than* the expected outcome of the hypothetical example. Person Z, being risk neutral, has preferences that are determined by the expected outcome of the treatment. Hence, 50 per cent intelligibility has a utility of .5 and is *equal to* the expected outcome of the hypothetical treatment example.

Utility Survey

As described above, the methods of utility assessment consist primarily of a series of value judgments in the context of a number of hypothetical treatments. In actual clinical decision-making situations, a treatment alternative under consideration can be evaluated by assigning utilities to its possible outcomes from the assessed utility scales. Of course, the utility scales must be defined over all the attributes that describe treatment outcomes.

The attributes included in the assessment of the degree of handicap that is to result from a treatment are cosmetic, speech, dental, hearing, and psychological. However, the costs of treatment to the family, insurance company, or state crippled children's program also constitute a handicap that must be considered in evaluating treatment alternatives. Of these possible handicapping conditions, dental and psychological attributes were not included in this study. The state-of-the-art in cleft lip and palate research is such that it is impossible to confirm or deny the hypothesis that children with clefts can be differentiated from those without on the basis of psychological characteristics (Wirls, 1971). In addition, evidence as to psychological handicaps on the part of parents is also lacking.

The dental handicap of cleft lip and palate is related to both speech and cosmetics. However, an individual has a wide range in which to compensate for dental abnormalities. The complexity of associating a level of handicap to a particular dental problem precludes it from consideration in this analysis (Moorrees, et al, 1971; Olin, 1971; Stahl, 1970).

For each of the remaining attributes, a scale that quantitatively described treatment outcomes had to be selected. For speech, intelligibility, as defined above, sufficed. However, a scale suitable for measuring cosmetic outcomes

required unique considerations. The measure had to describe the amount of facial disfigurement in terms that would be easily understood and easily communicated. Also, it had to be relatively unbiased by facial features unaffected by the cleft (the color of the eyes, for example). No such single measure existed in the surgical literature. Therefore, the approach used in this study was to select five children of approximately the same age with repaired complete unilateral clefts of the lip and palate. The children were selected to be representative of the range of possible cosmetic outcomes. In an attempt to eliminate biases that would be introduced by noncleft facial features, the mid-third of the face was extracted from each child's picture. An artist's sketch of a face, without the mid-third, was combined with these partial pictures to reconstruct five complete faces. These composite faces were then photographed in color so that the pictures of five different children became five different cosmetic results on a standardized face (Figure 2).

Of the five faces, Face "A" was selected as the preferred cosmetic result (the least degree of handicap), and Face "E" was selected as showing the greatest degree of handicap. By ranking the three remaining faces against these extremes, it was possible to determine utility values for a range of surgical results.

Hearing problems associated with clefts are usually measured in terms of decibel loss. Although decibel loss is a useful and valid measure, not many people can fully appreciate the impact of progressively greater amounts of loss. Thus, the only scale values used in this study for this attribute were 0 (a hearing aid needed) and 1 (no hearing aid needed).

The financial attribute is, of course, scaled in dollars. But those dollars could be costs to the child's family, income to the physician or hospital, or costs to insurers or crippled children's programs. Utilities would have to be independently assessed from representatives of each group. However, in the setting where the study was carried out, two factors combined to make this approach unworkable. First, the amount of money charged by a particular clinician for a single case is very small in relation to that clinician's overall income (i.e., while total payments can be large, they are distributed among several clinicians). Second, the extent to which third parties will pay for care and the charges themselves are inextricably related to the amounts that the family can pay. Hence, all utilities are conditional on the utilities of the family. For this reason, only a single dollar cost was considered for the financial attribute and all assessments were made in terms of the utilities of the child's family.

The evaluation of alternative treatments for cleft lip and palate must be made on the basis of the habilitation of the whole child and not on the basis of a check list of attributes. Evaluation, therefore, requires a scale over all attributes that properly weighs their interdependencies. Keeney (1974) has shown mathematically that, if certain assumptions can be made, the utility of a set of attributes can be expressed as the sum or the product of the utilities of the individual attributes coupled with some appropriate scaling constants.

The two assumptions needed are utility independence and pairwise preferential independence. Utility independence of an attribute means that a person feels that the utility of a treatment which involves only that attribute is independent of

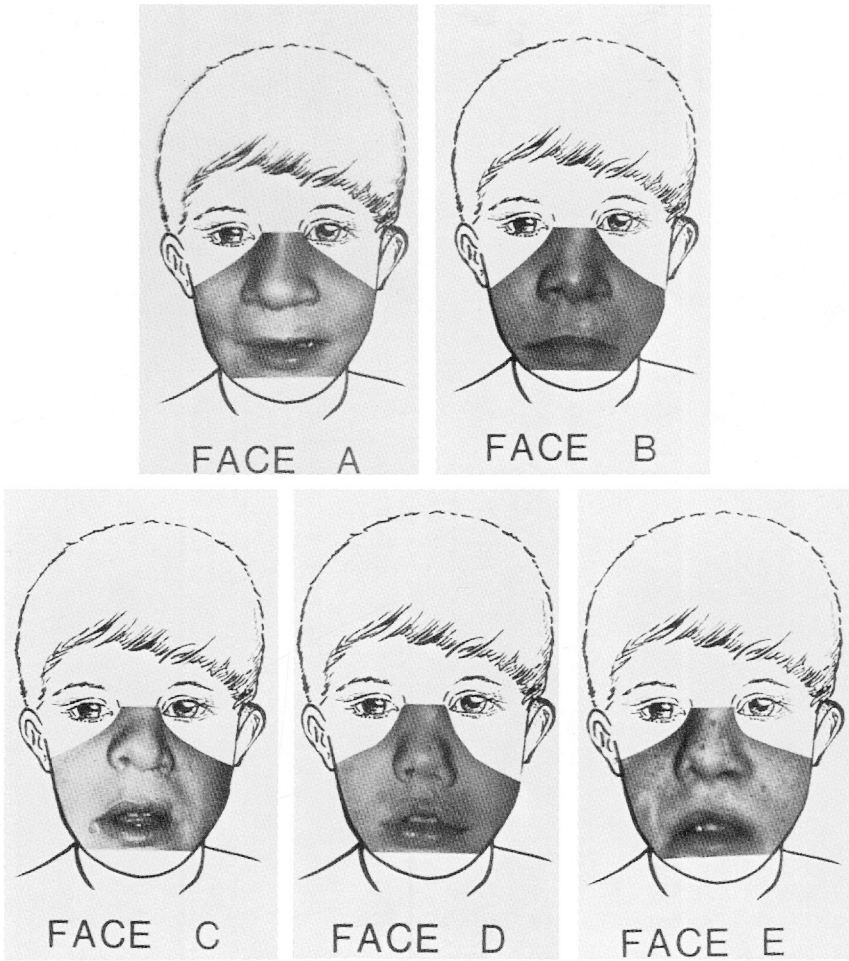

FIGURE 2. The five standardized faces depicting a range of cosmetic disfigurement after surgery. The faces were photographed in color for the utility assessments.

the levels of the other attributes. Pairwise preferential independence means that a person feels that the amount one would be willing to substitute one attribute for another is independent of the remaining attributes. In this study, it was hypothesized that most people would feel that treatment costs would be utility independent of speech, cosmetics, and hearing and that each of the attributes paired with costs would be preferentially independent of the remaining attributes.

A questionnaire was designed to assess the utility functions for each of the attributes selected to describe cleft lip and palate treatment outcomes. The questionnaire was also designed to test the validity of the utility independence and the pairwise preferential independence assumptions needed to formulate the multiattribute utility function.

Prior to its mailing, the questionnaire underwent careful testing. A selected number of clinicians and non-clinicians were asked to read each question aloud

and to give their own interpretation of what they thought the question asked. It was not until there was complete agreement between what they thought the questions asked and what was intended that the questionnaire reached final form.

Results

Responses were received from 89 individuals representing thirteen clinical specialties from cleft lip and palate teams at 17 of the 34 facilities contacted. Thirty members of families of cleft lip and palate children also responded to the questionnaire (Table 2). The questionnaires were not distributed directly to clinicians or to families, but rather to the facilities themselves. The responses represented nearly 20 per cent of the total number of questionnaires distributed. The survey and the responses to it were not intended to be random and so do not constitute a random sampling of either clinicians or families. The purpose of the survey was to determine the applicability of utility theory to cleft lip and palate treatment decision making and to see if differences among assessed utilities could be determined for clinicians and family members grouped according to their clinical specialty or the facility with which they were associated. Inasmuch as the selection of respondents was not controlled, the non-response rate is unknown. Hence, the interpretation of the results as being representative of all clinicians or families associated with cleft lip and palate must be considered with appropriate caution.

TABLE 2. Responses by clinical specialty and family membership.

respondents	*number of responses*
Clinical specialty	
Plastic surgery	17
Orthodontics	14
Speech pathology	11
Otolaryngology	7
Pediatrics	8
Prosthodontics	7
Social Work	1
Pedodontics	5
Audiology	2
Psychology	2
General dentistry	6
Nursing	4
Psychiatry	2
Unspecified	3
TOTAL	89
Family membership	
Mother	14
Father	9
Mother & father together	4
Patient	2
Other relative	1
Total	30

The answer sheet was designed to permit two sets of responses depending on the sex of the child with the cleft. Early in the development of the questionnaire, some clinicians advanced the idea that acceptable tradeoffs between speech and cosmetics might depend on the child's sex. The thinking was that cosmetics should be emphasized over speech for a girl and the reverse for a boy. One surgeon suggested that the boy could always grow a moustache to cover a poor cosmetic outcome. Also, the concept that girls needed to look pretty whereas boys needed to speak well to succeed in life was also advanced. These ideas met with varying degrees of amusement and disdain among other clinicians contacted. Nonetheless, the questionnaire permitted the expression of these notions.

The results of the utility survey indicated that only 13 (11 per cent) of those responding to the questionnaire varied their answers according to the sex of the child. No significant differences were found between the utilities of this group and those of the other respondents. If a child's sex does affect the utilities of treatment outcomes in a systematic way, it was not evidenced in this study.

UTILITY FUNCTIONS FOR TREATMENT COSTS AND MONETARY GAIN. The utility functions were assessed over possible treatment costs and for a range of values that represented monetary gain (such as from winning a lottery). Systematic differences in preferences and attitudes toward risk among team members of a facility and the members of patients' families treated at that facility were not statistically significant (Table 3). Only seven of the 17 responding facilities could be analyzed in this regard because of limited data from the other facilities.

TABLE 3. Attitudes toward risk (Selected Facilities).

facility	*monetary gain*			*monetary loss (cost)*		
	A	*N*	*S*	*A*	*N*	*S*
A (C)*	4	6	0	0	2	8
(F)*	0	2	2	0	1	4
B (C)	0	6	4	1	3	5
(F)	0	2	0	0	0	2
C (C)	4	3	2	0	1	8
(F)	1	0	0	0	0	1
D (C)	1	5	0	0	0	5
(F)	0	1	1	0	1	1
E (C)	5	5	3	0	5	7
(F)	0	1	3	0	1	3
F (C)	2	2	2	0	0	8
G (C)	1	4	0	0	2	3
(F)	0	2	2	1	0	3
Total (C)	17	31	11	1	13	44
Total (F)	1	8	8	1	3	14
Total for all facilities						
(C)	22	36	20	1	14	62
(F)	6	8	12	1	3	18

* (C) = Clinical Staff; (F) = Family Member; A = Risk Averse; N = Risk Neutral; S = Risk Seeking.

Comparisons between certain of the major clinical specialties and family membership (Table 4) were also statistically insignificant. It is interesting to note that there was an almost even split among respondents who were risk seeking, risk neutral, and risk averse in their utility functions for monetary gain. However, over 80 per cent of the respondents were risk seeking in their utility functions for costs.

A possible interpretation of this type of attitude toward risk is that people try to conserve resources. That is, the payments that they would expect to make in insurance premiums reach a limit beyond which additional payments would be as devastating as the costs of the hypothetical treatment.

UTILITY FUNCTIONS FOR SPEECH. Since the system used for measuring speech is the per cent of words from a sample of speech found to be intelligible, comparisons of utility functions for speech, like those for treatment costs, can be based on their shape. Concave utility functions, implying risk aversion, were reported by 55 per cent of those responding to the questionnaire. Utility functions for 30 percent were linear (risk neutral), and only 15 per cent had convex utility functions (risk seeking). Most of the linear functions included segments that were concave. Comparisons of respondents, grouped according to clinical specialty, family membership, or affiliation with a particular treatment center, yielded no statistically significant differences.

UTILITY FUNCTIONS FOR COSMETICS. These utilities were assessed for particular cosmetic results that were arbitrarily selected to represent a range of treatment outcomes. Since there was no measurable scale for each result, it was not possible to compare attitudes toward risk as they are expressed in the shapes of the assessed utility functions.

MULTIATTRIBUTE UTILITY FUNCTIONS. As noted above, the formulation of an approach to treatment evaluation must be more than a list of values that describe

TABLE 4. Attitudes toward risk (by selected clinical specialty and family membership).

respondents	*monetary gain*			*monetary loss (cost)*		
	A	*N*	*S*	*A*	*N*	*S*
Plastic surgery	4	10	2	1	5	8
Orthodontics	1	10	3	0	2	11
Speech pathology	3	3	4	0	1	9
Otolaryngology	3	1	1	0	0	5
Pediatrics	3	5	0	0	0	8
Prosthodontics	1	2	0	0	1	5
Pedodontics	2	2	1	0	1	4
General dentistry	1	2	2	0	2	4
Nursing	1	1	2	0	0	4
Total	19	36	15	1	12	58
Mother	3	3	6	1	1	9
Father	2	2	4	0	2	5
Mother & father together	0	3	1	0	0	3
Total	5	8	11	1	3	17

A = Risk Averse; N = Risk Neutral; S = Risk Seeking.

the level of handicap of each attribute individually. The individual attribute utility functions need to be aggregated in such a way as to represent an index of the overall habilitative status of the patient. The assumptions of preferential and utility independence enable the construction of a multiattribute utility function, defined over all the attributes, that meets these needs.

By design, it was possible to determine the validity of these assumptions for each attribute and respondent. It was determined that 60 per cent of those responding considered monetary expense to be utility independent of the other attributes. 71 per cent, 72 per cent and 75 per cent respectively considered the pairs money and cosmetics, money and speech, and money and hearing to be preferentially independent of the remaining attributes. No statistically significant systematic differences were found among the cleft palate team members for whom the assumptions were valid when analyzed according to facility affiliation. Also, differences in attitudes with regard to the utility independence of treatment costs among specialists or family members were not significant. However, for family members and selected clinical specialists, some differences were significant (Table 5). Significantly fewer fathers than mothers ($p < .05$), pediatricians ($p < .025$), and plastic surgeons ($p < .01$) felt that speech (paired with cost) was preferentially independent of the other attributes. Also, the proportion of speech pathologists for whom the pairwise preferential independence of speech and cost was valid was significantly less than either pediatricians or plastic surgeons ($p < .05$). Fathers accounted for all of the statistically significant differences among the respondents for whom the pairwise preferential independence of hearing and cost was valid. Significantly more fathers than mothers ($p < .05$), pediatricians ($p < .025$), and otolaryngologists ($p < .05$) felt that hearing paired with costs should not be preferentially independent of the other attributes.

A possible rationale for these differences could be that fathers felt cosmetics to be so important that the acceptability of any expenditure for speech or hearing had to depend on the final cosmetic outcome. For example, if the cosmetic result was very poor, then one might be willing to spend as much as necessary for the opportunity to improve speech or hearing. However, if the cosmetic result was

TABLE 5. Valid independence assumptions by selected clinical specialties and family members.

respondents	*number of responses*	*utility independence of costs*	*preferential independence of cost paired with*		
			cosmetics	*speech*	*hearing*
Clinical staff					
Plastic surgeon	17	7	13	15	12
Orthodontist	14	9	13	10	10
Speech pathologist	11	6	6	6	8
Otolaryngologist	7	4	6	6	7
Pediatrician	8	6	7	8	8
Prosthodontist	7	5	5	4	5
Family members					
Mother	14	7	10	11	11
Father	9	5	5	3	3

excellent, then one might be of the persuasion that expenditures to improve either speech or hearing should be more modest. Of course, this is conjecture, and all that can be said is that for fathers (and some speech pathologists) the utility of trading-off money for speech or hearing is dependent on the other attributes.

THE RELATIVE VALUE OF THE ATTRIBUTES. In arriving at a multiattribute utility function, each attribute is assigned a weight which may be loosely interpreted as an expression of the relative importance or value of each type of handicap associated with cleft lip and palate. These weights are calculated from the questionnaire responses as to how much of one attribute one would be willing to sacrifice for a measured improvement in another attribute. That is, by assessing how much degredation in speech quality one would find acceptable for a measured improvement in hearing ability, one can calculate the relative values of speech and hearing handicaps.

Information was sufficiently complete for 100 of the 119 respondents to permit the calculation of attribute weights. Data from 57 per cent indicated that the relative weight for speech was greater than that for cosmetics (Table 6). The relationship between the relative weights for costs and hearing was the same for 95 per cent of the responses analyzed. When the responses were grouped according to family membership and clinical specialty, the results showed that significantly more fathers than either mothers or plastic surgeons gave speech a higher weight than cosmetics. Comparing a facility which regularly uses presurgical orthopedics with two which do not, significantly more clinicians at the center using presurgical orthopedics gave a greater weight to cosmetics, while the majority of clinicians using purely surgical intervention gave the higher weight to speech.

In almost all cases, the values given to the scaling constants for speech and cosmetics were very close. The values given to the scaling constants for treatment costs and hearing were also close, but a fairly wide margin separated the speech and cosmetic scaling constants from them. Curiously, in over 95 per cent of responses analyzed, cost was given a higher weight than hearing loss. However,

TABLE 6. Orderings of scaling constants.

respondents	*ordering*		
	$k_1 > k_2 > k_3 > k_4$	$k_2 > k_1 > k_3 > k_4$	*other*
Facility "X" using presurgical orthopedics	5	1	0
Facility "Y" using purely surgical intervention	2	4	0
Facility "Z" using purely surgical intervention	3	11	0
Plastic surgeons	7	9	0
Mothers	8	6	0
Fathers	0	9	0
Total specialists	28	39	4
Total family members	10	18	1

k_1 = Cosmetics; k_2 = Speech; k_3 = Monetary Expense (Cost); k_4 = Hearing.

this may be an artifact of the questionnaire since it was stated that hearing loss is almost always treatable by the use of a hearing aid; and hearing aids are rarely required by cleft patients.

Discussion

The results of the utility survey indicate that some significant differences do exist among clinicians grouped according to their specialty or the facility to which they are affiliated. These differences suggest that variations in treatment decision making can, at least partially, be ascribed to variations in how the degree of handicap that is to result from treatment is perceived. That is, two groups or teams of clinicians may choose different "superior" treatments not because they differ in their clinical estimate of what the treatment outcomes might be but because they place different values on the amount of handicap that is to result from that treatment.

A good example of this effect can be drawn from the observed differences in utilities between a team regularly using presurgical orthopedics and a team not using presurgical orthopedics. Since very little data exist from which to determine the probabilities of the possible cosmetic, speech, hearing, and cost outcomes for these two approaches to treatment (Krischer, et al, 1975) subjective estimates had to be obtained from clinicians at each facility (Krischer, 1974). Using these probabilities and the utilities of a clinician at a facility using presurgical orthopedics, it was found that the "superior" treatment choice was, in fact, presurgical orthopedics. However, using the utilities of a clinician at a facility not using presurgical orthopedics the "superior" treatment was found not to be presurgical orthopedics. On this basis, the resolution of the controversy surrounding the use of presurgical orthopedics might better be approached by investigating why some treatment outcomes, as documented by the resulting degree of handicap, appear to be "better," or at least more acceptable, to some clinicians than to others. The answer may lie in the subjective nature of the information on which decision making was based.

Most of the differences found in the results of the utility survey were centered on the relative values of the several attributes that describe treatment outcomes. Fathers of cleft lip and palate children tended to be the single group that accounted for the majority of the observed differences. This seems to suggest that fathers do not view the habilitative goals of treatment programs in the same way as do either mothers or the team members. That mothers seem to have the same value structure as the teams which are providing services to their children may reflect the closer relationship a mother has with her child's treatment program. However, these conclusions are only tentative since certain biases could have been introduced in the selection of the families that participated in the utility survey. Also, it is conceivable that the utility structure of the clinicians is imparted to the family through the normal course of the child's treatment and that fathers tended to embrace the values of the clinicians to a lesser extent than mothers.

The assumptions that were needed to formulate a multiattribute utility function that would be a measure of the habilitative status before and after

treatment appeared to be valid for the majority of those responding to the questionnaire. Hence, utility theory is a reasonable approach both to the documentation of treatment outcomes in terms of the perceived degree of handicap and to the evaluation of alternative therapeutic procedures.

The specific results presented here must be considered in the context of the very difficult methods of utility assessment employed. In most applications, utility functions are assessed by personal interview. The questionnaire approach used in this study presented a number of difficulties. Principal among them were the phrasing of key questions and the difficulty in expressing the concept of indifference between a treatment with a certain outcome and an alternative with an uncertain outcome. Another problem encountered was the tendency for respondents to confuse a hypothetical treatment outcome with the utility of that outcome. This was especially true of questions asking about the interdependencies of the utilities of the individual attributes. Several respondents commented that treatments which altered one attribute almost always affected another. These comments are indicative of the confusion that existed between the probabilistic dependencies among attributes and the interdependencies of their utilities. Had the utility assessment been by personal interview rather than by questionnaire, these problems could have been resolved.

Utility assessment would appear to have considerable potential for treatment planning. Differences that could make some treatment decision unacceptable could be identified early in a program and corrective action taken so as to not jeopardize the continuity of care. Hence, incorporating utilities into the decision making process should allow for a more effective utilization of scarce and expensive medical resources which could be individually tailored to the perceived habilitative needs of the child.

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References

BETAQUE, N. E., *Utility Theory Applied to Medical Diagnosis and Treatment,* unpublished Master's Thesis, Massachusetts Institute of Technology, Cambridge, Massachusetts, (1969).

FORST, B. E., A Decision-Theoretic Approach to Medical Diagnosis and Treatment, Professional Paper No. 52, Center for Naval Analyses, Arlington, Virginia (November, 1970). Presentation, Eleventh American Meeting Institute of Management Sciences, Los Angeles, California, (October, 1970).

GIAUQUE W. C., *Prevention and Treatment of Streptococcal Sore Throat and Rheumatic Fever—A Decision Theoretic Approach,* unpublished D.B.A. Thesis, Harvard Business School, Harvard University, Boston, Massachusetts, (1972).

GINSBERG, A. S., Decision Analysis in Clinical Patient Management with an Application to the Pleural Effusion Syndrome, R-751-RC/NCM Santa Monica: The Rand Corporation, (1971).

KEENEY R. L., A Decision Analysis with Multiple Objectives: The Mexico City Airport, *The Bell Journal of Economics and Management Science, 4,* 101–117 (1973).

KEENEY, R. L., Multiplicative Utility Functions, *Operations Research 22,* 22–34 (1974).

KOEPP-BAKER, H., The Cleft Palate Team, in Grabb, W. C., Rosenstein, S. W., and Bzoch, K. R. (eds.), *Cleft Lip and Palate,* Boston: Little, Brown and Company, (1971).

KRISCHER, J. P., *An Analysis of Patient Management Decisions as Applied to Cleft Palate,*

unpublished Ph.D. Thesis, Harvard University, (1974). (Also, TR 12-74, Center for Research in Computing Technology, Harvard University, Cambridge, Massachusetts, 1974).

KRISCHER, J. P., O'DONNELL J. P., and SHIERE F., Changing Cleft Widths: A Problem Revisited, *Amer J of Ortho, 67,* 647–659 (1975).

MOORREES, C. F. A., BURSTONE, C. J., CHRISTIANSEN, R. L., HIXON, E. H. and WEINSTEIN, S. Research Related to Malocclusion, *Amer J of Ortho, 59,* 1–8 (1971).

OLIN W. H., Orthodontics, in Grabb, W. C., Rosenstein, S. W., and Bzoch, K. R. (eds.), *Cleft Lip and Palate,* Boston: Little, Brown and Company, (1971).

PLISKIN J. S., *The Management of Patients with End-Stage Renal Failure: A Decision Theoretic Approach,* unpublished Ph.D. Thesis, Harvard University, Cambridge, Massachusetts, (1974).

PRUZANSKY S., Description, Classification and Analysis of Unoperated Clefts of the Lip and Palate, *Amer J of Ortho, 39,* 590–611 (1953).

SPRIESTERSBACH, D. C., DICKSON, D. R., FRASER, F. C., HOROWITZ, S. L., MCWILLIAMS, B. J., PARADISE, J. L. and RANDALL, P., Clinical Research in Cleft Lip and Cleft Palate: The State of the Art, *Cleft Palate J. 10,* 113–165 (1973).

STAHL S. S., The Role of Occlusion in the Etiology and Therapy of Periodontal Disease, *Angle Orthodontist, 40,* 347–352 (1970).

SUBTELNY J. D., VAN HATTUM R. J. and MYERS B. B., Ratings and Measures of Cleft Palate Speech, *Cleft Palate J, 9,* 18–27 (1972).

SWALM R. O., Utility Theory-Insights Into Risk Taking, *Harvard Business Review,* 123–136, (November–December, 1966).

VON NEUMANN J. and MORGENSTERN O., *Theory of Games and Economic Behavior,* 2nd edition, Princeton: Princeton University Press, (1947).

WIRLS C. J., Psychosocial Aspects of Cleft Lip and Palate, in Grabb, W. C., Rosenstein, S. W., and Bzoch, K. R. (eds.), *Cleft Lip and Palate,* Boston: Little, Brown and Company, (1971).