A Psychosocial Study of Icelandic Individuals with Cleft Lip or Cleft Lip and Palate

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Questionnaires were sent to 63 Icelandic individuals with cleft lip and cleft lip and palate who had undergone surgical repair of those defects. The questions focused on the following areas: (1) social characteristics, (2) clinical characteristics, (3) attitudes toward consequences of clefts, and (4) attitudes toward appearance and treatment. The CL and CLP groups were compared, as were females and males. A comparison group was used with respect to two social factors. The data indicated relatively good psychosocial adjustment. However, fewer individuals were married or living together in the cleft group than in the comparison group. The subjects did not perceive that cleft lip or palate had influenced their lives to a great extent, yet females seemed to be more self-conscious than males in respect to how others viewed their appearance. Overall, the subjects were satisfied with the treatment and the members of the treatment team. However, their expectations exceeded the actual outcome of surgery. Suggestions are offered for future research.

In spite of great advances made in the management of patients with cleft lip (CL) and cleft lip and palate (CLP) during the last decades, these individuals have to carry the stigma of their deformity for a part of their lives. Those most severely deformed have some of the stigma their whole lives, even though they get the best expert treatment.

CL and CLP are among the most common congenital deformities with external manifestations. Möller (1971) found in an epidemiologic study that the frequency of clefts in Iceland was 2.51 per thousand births (individuals born 1954 to 1966). Among North American Caucasian children, the estimated annual incidence is about 0.9 per thousand live births (Heller et al, 1981). This indicates that in every society there is a considerable number of adult individuals with some of the stigmata of this congenital deformity, individuals who have gone through some or all stages of current treatment.

Research on the cleft lip and palate problem has been pursued by many groups of scientists in the fields of medicine, dentistry, epidemiology, and genetics. Until recently, however, little was found in the CLP literature on the psychosocial effects of facial anomaly and the speech and hearing defects that often accompany CLP. External appearance is very important in our present day society, and deviations from the normal such as CLP and other facial and craniofacial deformities can have profound effects on the individual and his surroundings (Clifford, 1973; Hanus et al, 1981). Since facial anomaly affects not only the individual but his family and peers as well, extensive psychosocial influence can be predicted. It is not only the individual's own perception of the condition that contributes to the effects, but the reactions of all the people in his or her environment as well (Clifford, 1973; Edwards and Watson, 1980).

Several studies during the past decade have provided some insights into the psychological adjustment of individuals with CLP. For example, a high degree of dissatisfaction and self-consciousness regarding appearance was reported by Heller et al (1981) and Birch and Lindsay (1971); a low rating for body-satisfaction was reported by Clifford et al (1972), and evidence that individuals with CLP appear to be observers rather than active participants in social interactions and feel neglected because of their handicap was reported by VanDemark and VanDemark (1970). Many patients report that they have ex-

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perienced problems in relationships with the opposite sex (Birch and Lindsey, 1971; Heller et al, 1981), and they seem to marry at a later age and at a lower rate compared to siblings and control groups (Heller et al, 1981; McWilliams and Paradise, 1973; Peter and Chinsky, 1974a). The majority has experienced problems in relationships with peers and a restriction of participation in social activities; for example, they have been teased about their defects during their school years (Birch and Lindsey, 1971). On the whole they seem to show a prolonged dependence on the family and lower levels of social integration (Heller et al, 1981; Peter et al, 1975; Spriesterbach, 1973). A large proportion has expressed the need for more counseling and supportive services (Heller et al, 1981). Yet they seem to have achieved educational levels similar to siblings and control groups (McWilliams and Paradise, 1973; Peter and Chinsky, 1974b). Although the general picture is negative, some researchers have not found differences between individuals with CLP and normal subjects with regard to the above (for a review, see Edwards and Watson, 1980).

PURPOSE OF THE STUDY

In plastic surgery, treatment is often difficult to evaluate. Evaluations of operations in terms of surgeons' satisfaction is incomplete. The goal that every plastic surgeon must try to reach with treatment—patient satisfaction and good social adjustment—is difficult to achieve. The present study is the first attempt in Iceland to evaluate the outcome of treatment of patients with CL and CLP with the focus on the patients' point of view. Our immediate interest was how these individuals view themselves in society and how they look upon services they have received to normalize their form and function.

Iceland is relatively small; we were therefore optimistic about reaching all individuals with CL and CLP in certain age groups. Accordingly, demographic variables were among our interests.

Individuals born with cleft palate only were excluded from the research group, since the research focused on how people experience being born with clefts and associated facial deformities. Two groups were compared: those born with CL only and those born with both cleft lip and palate. Patients with CLP usually have more facial deformity than patients with CL and are more likely to have concomitant speech and hearing handicaps. The research included sex as a separate variable in order to detect possible differences in attitudes toward the cleft consequences between males and females.

METHOD

The initial pool of subjects consisted of 79 individuals, 23 females and 56 males, namely all those individuals born from 1936 through 1960 who were diagnosed as presenting cleft lip or cleft lip and palate and who were operated on at least once during the years 1936 to 1981 in the University Hospital in Iceland. Ten of those could not be located, so the questionnaires were sent to the remaining 69 individuals (46 males, 23 females). Sixty-three questionnaires (91 percent) were returned.

The questionnaire included mainly structured questions that dealt with the following topics: (1) social characteristics of individuals with CL or CLP, (2) clinical characteristics, (3) attitudes toward the consequences of clefts, and (4) attitudes toward appearance and treatment.

Descriptive statistics were obtained for the social and clinical characteristics portions of the questionnaire. Two questions in the social characteristics portion (regarding education and marital status) had been answered by members of a group of 326 individuals in a different study. Their responses were used for comparison purposes. The members of this group of 163 males and 163 females were normal individuals who ranged in age from 25 through 50 years. They were recruited by a random selection from this age range of the whole Icelandic population (for the purpose of studying leisure-time activities).

Subjects were instructed to use a five-point scale to respond to questions about attitudes (topics 3 and 4 above). Items were structured to permit expressions ranging from positive to negative adjustment: very little versus very much, poorly versus well, rarely versus often. Except for items regarding operations, a scale value of 1 was assigned to the most positive response and a value of 5 was assigned to the most negative response (e.g., question 10: people stare *very often*). A 2 (CL, CLP) × 2 (males, females) analysis of variance was performed on the data from each of the 22 attitude questions. Since the attitude questions emphasized perception of the cleft consequences, no control group was included.

RESULTS

Social Characteristics

Of the 63 individuals who returned the questionnaires, 26 (41.3 percent) were born with cleft lip only and 37 (58.7 percent) with both cleft lip and palate. There were more males (63.5 percent, N = 40) than females (36.5 percent, N = 23) which reflects the higher incidence of males in the CL and CLP population (Heller et al, 1981; Möller, 1971). Most respondents (49.2 percent) were between 21 and 25 years of age. Of the individuals with cleft, 50.8 percent (N = 31) were married or living unwed with a person of the opposite sex, 31.1 percent (N = 19) were still living with parents, and the rest (18 percent) were living either alone or with friends.

Fifty percent of the males in the cleft group were or had been married or living unwed with a female. This compared with 87.9 percent of the males in the comparison group. More females (60.9 percent) than males (50 percent) in the cleft group were married or living unmarried with a partner. Nevertheless, the proportion of married females in the cleft group (60.9 percent) was lower than the proportion of married females in the comparison group (89 percent).

Table 1 shows the highest educational levels of the subjects. Level was defined as the last examination taken from a particular school. There was no significant difference between the cleft group and the comparison group. Chi-square analysis on sex and cleft groups revealed no significant differences regarding education, i.e., the educational levels of the patients were similar, whether they had CL or CLP, or whether they were male or female.

Clinical Characteristics

Eleven females and 15 males (41.3 percent of the subjects with clefts) were born with cleft lip only; and 12 females and 25 males (58.7 percent) were born with CLP. Approximately half of the cleft group had a family history of clefting (46 percent). Most of the subjects (57.4 percent) had undergone at least four operations in connection with the clefts. Otitis media afflicted 40.3 percent of the subjects in childhood. Table 2 shows the proportion of the cleft group that has had various kinds of special treatments. In three areas an expected significant difference ($p \leq 0.05$) was detected with respect to diagnostic groups: (1) speech therapy, CLP = 40.5 percent vs CL = 7.6 percent; (2) hearing therapy, CLP = 11.3 percent vs CL = 0.0 percent; (3) dentures, CLP = 27 percent vs CL = 6.3 percent.

Attitudes Toward the Consequences of Clefts

Twelve questions were used to detect patient attitudes about the consequences of clefts (see Table 3). Each question was analyzed separately with respect to type of cleft and sex. The questions dealt with the following: (1) teased in school, (2) neglected in school, (3) educational progress influenced by cleft, (4) relationship with peers influenced by cleft, (5) relationship with the opposite sex influenced by cleft, (6) siblings' teasing, (7) self-confidence influenced by cleft, (8) confidence in public influenced by cleft, (9) shyness experienced because of cleft consequences, (10) people staring, (11) people asking questions about cleft consequences, and (12) people feeling sorry for CL or CLP subject. The questions were rated on a 1 to 5 scale as previously mentioned.

TABLE 1 Educational Levels of Cleft Lip or Cleft Lip and Palate Patients and of Comparison Group Members

	Cleft Group (%)	*Comparison Group (%)*
Elementary (6–8 yr)	20.7	22.1
Middle school (9–10 yr)	22.4	18.1
Occupational (11–12 yr)	12.1	12.3
Gymnasium* (14 yr)	13.8	4.6
University (≥17 yr)	6.9	10.2
Other	24.1	32.8

* equivalent to North American high school

On the whole the subjects' adjustment seems adequate in that mean ratings range from the middle to the positive end of the scale (see Table 3). Unfortunately, as a comparison group is missing, the study does not allow any comparison with normal subjects on these questions.

Analysis of variance on each of the questions revealed a significant difference with respect to sex on the question "people stare," where the females had higher means than the males (2.8 vs 1.96) ($F_{1,53} = 4.55$, $p \leq 0.038$), indicating that women more than men feel that other people stare. Marginal significance ($p < 0.10$) was achieved on the question "people ask questions" also with respect to sex ($F_{1,53} = 3.36$, $p \leq 0.072$), with women more than men feeling that people do ask questions about the cleft consequences (Means: 2.32 vs 1.69). Marginal significance was also achieved with respect to sex on the variable

TABLE 2 Percentages of Patients with Cleft Lip or Cleft Lip and Palate Who Received Associated Treatments

Treatment	*Patients Treated (%)*
Speech therapy	27.0
Hearing therapy	11.3
Psychological assistance	9.5
Orthodontic treatment	60.3
Other major dental treatment	33.3
Dentures	33.3
Operations abroad	17.5

TABLE 3 Mean Scale Values Reflecting Attitudes Toward the Consequences of Clefts

	Males		*Females*		*Value**	
Questions	*CL*	*CLP*	*CL*	*CLP*	*Sex*	*CLP*
Teased in school	1.92	2.04	1.73	2.82	NS	NS
Neglected in school	1.62	1.73	1.55	2.09	NS	NS
Educational progress affected	1.85	1.57	1.27	1.09	<0.09	NS
Relationship with peers	1.46	1.46	1.64	1.82	NS	NS
Relationship with opposite sex	2.31	2.75	1.91	2.82	<0.09	NS
Siblings' teasing	1.18	1.10	1.00	1.18	NS	NS
Self-confidence	2.45	2.81	3.00	3.36	NS	NS
Confidence in public	2.50	2.41	3.00	2.80	NS	NS
Shyness	2.77	3.04	2.82	3.73	NS	NS
People staring	1.92	2.00	2.64	2.91	<0.04	NS
People asking questions	1.69	1.68	2.45	2.18	<0.07	NS
People pitying	1.64	1.38	1.55	1.91	NS	NS

* NS = not significant

"relationship with the opposite sex influenced by cleft consequences" ($F_{1,55} = 3.0$, $p \leq 0.09$), with males reporting that the cleft consequences had had more of an influence on their relationship with the opposite sex than females reported (Means: 2.53 vs 2.37). (Remember that more females than males were married or living with a partner.) Marginal significance was again found on the variable "educational progress influenced by cleft" ($F_{1,54} = 2.93$, $p \leq 0.093$), with males reporting more of an influence than females (Means: 1.71 vs 1.18). The 2 × 2 analyses of variance revealed no significant difference with respect to symptom group on any of the questions.

Attitudes Toward Appearance and Treatment

The three questions regarding attitudes toward appearance (Table 4) were analyzed in the same way as previous attitude questions, that is, low values were more favorable than high values. The three items were: (1) appearance makes a difference, (2) appearance makes a difference for others, and (3) current appearance satisfaction.

The subjects seemed to be relatively satisfied with their appearance (question 3 above) (M = 3.65). On the other hand, they said that appearance was more important for others than for themselves (overall means: 3.37 [for question 2] vs 2.67 [for question 1]). Analysis of variance on the question whether appearance is important for others revealed a marginally significant difference between males (M = 3.05) and females (M = 3.69) ($F_{1,55} = 3.92$, $p \leq 0.05$), where females assign a greater value to the importance of appearance for others than do men. Analysis of variance also revealed a marginally significant interaction on question 1 dealing with how much difference appearance makes for themselves ($F_{1,55} = 3.2$, $p \leq 0.08$). Females with CL assigned the highest degree of importance to appearance compared to all others (M = 3.36), and females with CLP the lowest (M = 2.09); the reverse was true for males with CL (M = 2.54) and CLP (M = 2.71).

In order to investigate the attitudes toward treatment in general, six questions were included: (1) satisfaction with surgery, (2) expectations from surgery, (3) satisfaction with treatment, (4) satisfaction with members of the treatment team, (5) fulfillment of special wishes, and (6) effects of deformity on current life. Here low ratings reflect undesirable responses and high ratings desirable responses, except for the last item (effects of deformity on current life) where a low rating is desirable (little effect) and a high rating undesirable (much effect).

Overall, the subjects seem to be relatively satisfied with the treatment (M = 4.16), their operations (M = 4.06), and the members of the treatment team (M = 4.4). Subjects reported that most of the time their wishes, if any, had been fulfilled (M = 4.24). Nevertheless, the group as a whole seems to have had higher expectations regarding the outcome of the surgery than was actually realized (M = 3.35). Most subjects reported relatively little overall effects of the deformity on their current lives (M = 1.75). No significant differences were obtained with respect to sex and symptom groups on any of the above six questions.

DISCUSSION

In general, our data indicate that the individuals with cleft in Iceland are relatively well adjusted socially. In spite of this, there is a smaller proportion of the cleft group than of the comparison group married or living unmarried with a person of the opposite sex, which is in agreement with previous studies (Heller et al, 1981; McWilliams and Paradise, 1973; Peter and Chinsky, 1974a). However, the majority of subjects in the

TABLE 4 Mean Scale Values Reflecting Attitudes Toward Appearance and Treatment

	Males		*Females*		*Value*	
Questions	*CL*	*CLP*	*CL*	*CLP*	*Sex*	*Cleft*
Appearance is important for self	2.54	2.71	3.36	2.09	NS	NS
Appearance is important for others	3.31	2.79	3.55	3.82	<0.05	NS
Current appearance satisfactory	3.90	3.80	3.91	3.00	NS	NS
Satisfied with treatment	4.40	4.00	4.00	4.25	NS	NS
Satisfied with members of treatment team	4.60	3.95	4.55	4.50	NS	NS
Satisfied with surgery	4.31	3.75	4.18	4.00	NS	NS
Expectation from surgery met	2.91	3.29	3.73	3.45	NS	NS
Fulfillment of special wishes	4.00	4.20	4.36	4.38	NS	NS
Effect of deformity on current life	1.55	1.81	1.64	2.00	NS	NS

research group were a few years younger than in the comparison group; consequently, that might have influenced the results (comparison group: 21 to 45 years old and cleft group: 25 to 50 years old).

The results are in line with other studies with respect to education, i.e., that cleft individuals seem to achieve educational levels similar to others (McWilliams and Paradise, 1973; Peter and Chinsky, 1974b).

Addressing the attitudes toward the consequences of clefts, it may be concluded that the subjects do not seem to perceive that the cleft consequences have influenced their lives to a great extent. The relatively high expression of satisfaction is perhaps attributable to factors likely to be operating in a retrospective study like this one. Time "heals wounds," and we tend to push painful experiences of our childhood into the deeper recesses of the mind (Clifford et al, 1972; Edwards and Watson, 1980; Heller et al, 1981).

Since appearance is especially important to females in our western society, the sex differences that were detected in the present study could be explained by different socialization of the sexes. In general, females seem to be more self-conscious about their appearance, which is in line with what we would have expected.

The results of the study indicate that counseling or supportive services for individuals with cleft, as for other individuals with a chronic disorder, might be of value (see Goldberg et al, 1979; MacGregor, 1979; Stenhauer et al, 1974) because subjects generally expect better results from surgery than they receive.

One important advantage of the present study is that it is free from selection bias, which has been a problem in some previous studies (e.g., Clifford et al, 1972). Almost all patients with cleft lip or cleft lip and palate who have been treated in Iceland have undergone surgery in the University Hospital. This gives us a more representative sample, and in this respect Iceland offers great opportunities for research on patients with cleft lip and palate.

As mentioned above, one of the major flaws in previous studies is that they are retrospective. Future research needs to follow individuals from childhood into adulthood in order to evaluate the psychosocial influences of cleft lip and palate. As this is costly research, another approach might be to use younger age groups so that the events are closer in time, an approach causing less retrospection. The results might then be compared to the results of the present study.

An interesting sequel is that the study seemed to serve as a kind of motivation for 9 of the subjects (over 14 percent) to seek further treatment with the improvement of appearance in mind.

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