

Approaches to Craniofacial-Specific Quality of Life Assessment in Adolescents

TODD C. EDWARDS, PH.D.
DONALD L. PATRICK, PH.D.
TARI D. TOPOLSKI, PH.D.
CASSANDRA L. ASPINALL, M.S.W.
WENDY E. MOURADIAN, M.D.
MATTHEW L. SPELTZ, PH.D.

Objective: To ascertain the domains that adolescents aged 11 to 18 years with congenital and acquired craniofacial differences (CFDs) consider important to their quality of life (QoL) to create a craniofacial-specific module.

Design: Interviews and inductive qualitative methods were used to guide the development of a conceptual and measurement model of QoL among adolescents with CFDs.

Setting: The Craniofacial Center at Children's Hospital and Regional Medical Center in Seattle, Washington.

Patients, Participants: Thirty-three in-depth interviews with adolescents (aged 11 to 18 years), one young adult interview (age 19 years), 14 in-depth interviews with parents, one young adult focus group, one parent focus group, and one panel of researchers and clinical professionals working in the field.

Results: Using the qualitative methodology, grounded theory, seven domains that adolescents with CFDs perceive are important to having a good QoL were found. Six of the domains (coping, stigma and isolation, intimacy and trust, positive consequences, self-image, and negative emotions) comprised the Youth Quality of Life Instrument–Facial Differences module. One other domain, surgery, was a salient issue for many of the youth, but not all, so it was made into a separate module, the Youth Quality of Life Instrument–Craniofacial Surgery module. This module relates to the experience of surgery, outcomes of surgery, and preferences for future surgery.

Conclusions: Using an established qualitative methodology, two QoL modules specific to adolescents with CFDs were developed and are ready for psychometric validation. Potential uses of the instruments are discussed.

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The enhancement of quality of life (QoL) is commonly invoked as one of the justifications for craniofacial surgeries and other interventions in childhood and adolescence (Pope and

Dr. Edwards, Dr. Patrick, Dr. Topolski, Dr. Mouradian, and Dr. Speltz are with the University of Washington, and Ms. Aspinall, Dr. Mouradian, and Dr. Speltz are with the Children's Hospital and Regional Medical Center, Seattle, Washington.

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Address correspondence to: Todd C. Edwards, Ph.D., 146 North Canal Street, Suite 313, Seattle, Washington 98103-8652. E-mail toddce@u.washington.edu.

Speltz, 1997). Specific questions such as, “Will a successful result improve the patient's life?” are being considered in addition to the traditional concerns of functional and physiological benefits (Canady, 1995). Often, however, it is the physician or parent who makes the judgment regarding how surgery will improve QoL for the patient who is a minor.

Youth's voices are often translated through complex interactions among parents, surgeons, and other clinicians. Research has demonstrated, however, that a significant proportion of adolescents feel left out of treatment decisions in the craniofacial setting and that, when questioned, their satisfaction with clinical outcomes often do not correlate with parents who may be making the treatment decisions for them (Turner et al., 1997). Given the largely subjective nature of QoL questions, and the fact that it is the adolescent who undergoes treatment and has to live with its consequences, it is important to consider the adolescent's own perspective, desires, and expectations (Mouradian, 1999). This article presents the conceptual

framework, methodological considerations, and content-specific items in craniofacial-specific modules for measurement of adolescent QoL.

Assessing Quality of Life and Psychological Outcomes in Adolescence

Adolescence is a critical time for individuals with both congenital (Padwa et al., 1991) and acquired craniofacial differences (CFDs; McQuaid et al., 2000). Adolescents with CFDs encounter the typical developmental tasks of this period but must also struggle with the impact of their condition on appearance and sense of self. Adolescence is a time of heightened sensitivity to appearance and attractiveness and a time when identity issues become paramount. This period also marks the end of facial growth and associated surgeries and other essential craniofacial treatments. Key decisions concerning elective treatments are therefore often made at this time. These youth may hold unrealistic expectations for improvements with craniofacial surgeries or, conversely, refuse potentially beneficial interventions simply because they imply that the adolescent is in some way inadequate. Even youth undergoing dramatic changes with surgery may not experience an improved sense of self because of psychosocial problems rooted in years of prior negative experiences (Pillemer and Cook, 1989). For all these reasons, it is important for providers to understand adolescents' expectations of surgery (Tung and Kiyak, 1998).

Outcome research on children with congenital CFDs has focused primarily on provider-driven measures such as orthodontic, speech, and aesthetic outcomes (Shaw et al., 1992a, 1992b, 1996; Sandy et al., 1998). However, it has been shown that objective medical indicators do not reliably predict psychological adjustment in individuals with congenital craniofacial conditions (Robinson, 1997). Research on burn victims suggests frequent lack of correlation between severity of injury and psychological adjustment (Lansdown et al., 1991; Robert et al., 1998).

Psychological studies of youth with CFDs have also emphasized clinician-defined outcomes such as cognitive abilities and academic achievement (e.g., Richman and Ryan, 2003), behavioral adjustment (Pope and Ward, 1997b), and quality of parent-child and peer relationships (Tobiasen and Speltz, 1996; Kapp-Simon and McGuire, 1997; Pope and Ward, 1997a; Speltz et al., 1997). These studies demonstrated that adolescents with CFDs are at increased risk for problems in learning, behavior, and social competence. Long-term outcome studies of adults with congenital CFDs also report a negative impact on marital and financial/job status (Nash, 1995). Psychological studies that examined self-perception or self-esteem of youth with congenital CFDs have yielded inconsistent results, raising questions about the usefulness of these constructs for this population (Eiserman, 2001).

Because improvement of QoL is the justification for many craniofacial surgeries and other interventions, appropriate assessment tools should be employed with this purpose in mind.

In developing the craniofacial-specific QoL modules described in this article, we were guided by the World Health Organization (WHO) definition of QoL as people's "perceptions of their position in life in the context of the culture and value systems in which they live, and in relation to their goals, expectations, standards, and concerns" (WHO QoL Group, 1994; Bonomi et al., 2000). This definition dictates that youth themselves, rather than providers, define the important concepts and items, that the measure employ subjective self-report, and that the items be developmentally appropriate. This approach emphasizes a positive, "glass half-full" orientation toward health-enhancing aspects of life rather than the negative "glass half-empty" orientation of many assessments. It is hoped that the QoL paradigm with its positive assets approach will help identify sources of resilience and strategies of coping that are rarely seen in deficit-oriented approaches (Broder, 2001; Eiserman, 2001; Mouradian, 2001; Strauss, 2001).

Even when craniofacial treatments have clear goals in improving functional status, QoL outcome measures are needed to supplement the more technical and provider-driven measures. The subjective impact of changes in facial appearance, dental status, jaw relationships, speech intelligibility, and burden of treatments may be substantial. For those with acquired conditions, the subjective impacts of multiple surgeries and social interaction and peer relationships are needed to evaluate treatment with craniofacial reconstruction.

QoL outcome assessments and evidence-based methodologies are also needed to demonstrate to health policy makers and third-party payers the impact of CFDs and related treatments, especially those considered elective. Health insurers and managed care plans increasingly look to evidence-based research to support the effectiveness of often-expensive treatments (Clancy and Eisenberg, 1998; Nelson et al., 1998). Moreover, without evidence in support of efficacy, there are serious ethical objections to exposing patients, especially minors, to the risk of invasive surgeries. A better understanding of the subjective experiences of patients with CFDs and their families will benefit the adolescents facing these decisions, parents making decisions for their infants and young children, and clinicians involved in this field. From an ethical standpoint, adolescents must be involved in the decision-making process even if they cannot provide full informed consent, particularly when considering elective QoL interventions (see Mouradian, 1999, for a review of minors' decision making in the craniofacial context). Research has shown that a significant proportion of adolescents feel left out of treatment decisions in the craniofacial setting (Turner et al., 1997).

In summary, the foregoing review argues for special attention to the subjective experience of children and adolescents with CFDs. For methodological reasons, we chose to begin our study of these issues by interviewing adolescent patients capable of abstract thinking and self-reflection before grappling with the complexities of assessment and self-report in younger children.

METHODS

Data Collection

In developing the modules, the following data were collected: (1) 33 semistructured, in-depth interviews with adolescents aged 11 to 18 years with a range of congenital and acquired CFDs on how their CFDs affected their lives; (2) a subset of 15 in-depth interviews with parents of these adolescents (the first 15 parents who agreed to be interviewed); (3) one focus group with young adults ($n = 5$) with CFDs (aged 19 to 25 years) in which a moderator led a group discussion; and (4) one focus group with parents ($n = 8$) of adolescents with CFDs (included some interviewees). In addition, an advisory meeting was held with clinical and academic experts ($n = 10$) working in the field in which the newly developed items were evaluated. The interviews with the adolescents formed the primary basis for item generation, with the young adult focus group and parent interviews also generating items but serving primarily as confirmation checks that no major issues were missing (from their perspective). The parent focus group and the expert panel were used for the purpose of reducing the large item bank generated by the adolescent interviews to a manageable amount. The rationale for this mix of interviews and focus groups was to capitalize on the strengths of each format to maximize the amount of information gained from the different types of participants.

The adolescents who were interviewed were purposely selected to represent a broad range of craniofacial conditions: (1) acquired (burns, other trauma) ($n = 5$); (2) birth mark ($n = 1$); (3) branchial arch disorders ($n = 4$); (4) isolated or syndromic craniosynostoses ($n = 4$); (5) cleft lip, cleft palate, or both ($n = 15$); and (6) other diverse conditions ($n = 4$). These interviewees were evenly divided by sex ($n = 16$ boys, $n = 17$ girls) and age group ($n = 18$ aged 11 to 14 years, $n = 15$ aged 15 to 18 years), and they were Anglo ($n = 22$), Hispanic ($n = 6$), and Asian/Pacific Islander ($n = 5$). The main objective of this sampling approach was to articulate a diverse set of perspectives regarding adolescent QoL, rather than to obtain a representative sample per se (Strauss and Corbin, 1990).

Research participants were recruited through the Craniofacial Center of Children's Hospital and Regional Medical Center in Seattle and also through the University of Washington School of Dentistry. Participants were paid a small amount of money as a token of appreciation for their involvement in the study. The recruitment protocol was approved by the Institutional Review Board of Children's Hospital.

The adolescents were approached to participate in the research project as "expert informants" to help design a questionnaire to assess adolescents' thoughts and feelings about their QoL. Individual interviews were conducted by one of three members of the research team experienced in qualitative interviewing. We continued to recruit interviewees until what they were telling us became redundant with what previous interviewees had said and little new information was being gained.

Procedure

Adolescent interviewees were invited to generally discuss their lives in relation to others their age, including values, goals, and expectations. Specific probes were used to illuminate stage-salient contexts of adolescence including home, school, work, and community (Bronfenbrenner, 1979), and how they perceived their lives were affected by having a CFD. The other interview and focus group participants were asked similar questions with regard to their view of adolescent QoL in general and how they perceived it was affected by having a CFD.

In accordance with the WHO QoL definition cited above, we used the needs-based model to create two brief craniofacial-specific QoL modules to augment the Youth Quality of Life Instrument—Research Version generic instrument (Edwards et al., 2002; Patrick et al., 2002). The needs-based approach to development of QoL measures builds on functional status measurement and views QoL as the net result of a person's evaluation of how much their needs have been met including their evaluation of functional status and interaction with the environment. It is based on Maslow's (1954) needs hierarchy and was the basis for the WHO-sponsored measure of QoL (Skevington, 2002). Also pioneering in this approach was Jan Hornquist from Scandinavia, and in general the needs-based approach has been a current in Scandinavian outcomes assessment for a number of years. The approach is discussed in more detail in articles by Hunt, McKenna, and others in reference to other instrument development (Hunt and McKenna, 1992; McKenna et al., 2001). Thus, the items comprising the instruments described in this article were selected to represent the areas of greatest salience as identified primarily by the adolescents with CFDs themselves.

An item pool was generated from qualitative interviews and focus groups with adolescents with CFDs and input from clinicians working in the craniofacial field. The parent interviews were used to supplement the information from the adolescents themselves. Normally, existing measures would have been consulted for this purpose as well, but as discussed above, the instruments available in the field are focused primarily on functional limitations, anatomical variables, and psychosocial maladjustment. We found no existing instruments that reflected a needs-level approach to the assessment of QoL.

Data Analysis and Item Development

A "grounded theory" approach guided data analysis (Glaser and Strauss, 1967). Grounded theory is derived from the sociological theory of symbolic interactionism (Blumer, 1969) and is used to model phenomena about which little is known, in this case the QoL of adolescents with CFDs. It is an inductive process approach, with an emphasis on social dynamics. The basic tenet of symbolic interactionism is that people construct meanings about their lives on the basis of interactions they have with other people and the world at large.

The investigators, previously experienced in this approach

(Edwards et al., 2002) worked with the transcribed interviews to code relevant QoL issues and to write items based as closely as possible on the views and language of the adolescents themselves. Data-coding strategies included open coding; assignment of codes to the text based on words or phrases that captured meaning in the data; axial coding, comparing open codes with each other to create relevant categories; and selective coding, using frequently occurring axial codes to create core categories, or conceptual model domains (for a full explanation of these coding processes, see Strauss and Corbin, 1990).

Over an 18-month period, 13 steps were used in the coding and analysis process: (1) The interviews were audiorecorded and transcribed; (2) each transcription was checked for accuracy; (3) at least two team members selected QoL-relevant text from each transcription; (4) the selected text was transferred to a spreadsheet along with its interview number; (5) team members began coding text and generated a long list of codes (open coding); (6) the long list of codes was consolidated into categories (axial coding); (7) all selected text was coded with the axial codes; (8) the selected text was sorted by axial code and further consolidated into core domains (selective coding); (9) draft items were written based on the text comprising the core domains; (10) the number of draft items was reduced based on investigator judgment of the importance of items; (11) draft items were “wordsmithed,” maintaining original language as much as possible; (12) a reduced list of draft items was presented to a group of four youth with CFDs, a panel of 15 researchers and clinicians, and a group of six parents for further reduction; and (13) a final set of items was cognitively debriefed with adolescents and prepared for field testing. Cognitive debriefing is a method by which individuals assess the relevance, importance, and comprehension of the content of measures (Jabine et al., 1984; Fowler, 1993). The four adolescents with CFDs who participated in step 12 above also completed the draft instrument and afterward were asked to “think aloud” about how they interpreted each item and how they chose a response. They were also asked to identify awkward or unclear wording and to evaluate whether any important issues were not included.

Two types of items, perceptual and contextual, were developed for the facial differences and facial surgery modules. Perceptual items measure those aspects of QoL known only to the adolescent respondent, which cannot be observed by others. Contextual items are self-reported but are potentially verifiable or observable by others. Both types of items are considered important for assessing QoL (Cummins, 1997; Wallander et al., 2001).

RESULTS

The original number of items generated was 845, all perceptual, distributed across seven categories: (1) coping, (2) intimacy/trust, (3) negative emotions, (4) positive consequences, (5) self-image, (6) stigma/isolation, and (7) surgery. Although some adolescent interviewees discussed functional issues, they were not included as a separate instrument domain because they are covered adequately by pre-existing instruments (as

discussed above). In step 10 above, each investigator nominated what he or she considered the best items. This resulted in 446 items remaining in the pool.

The principal investigator and project manager then evaluated each of these items and retained the best QoL items based on the following criteria: (1) the item evaluated a quality (perception/sensation/feeling), (2) the item represented an area of importance to people with the condition, (3) the item was in the language of the people with the condition and the item was translatable conceptually, (4) the item was likely to change with successful treatment of the condition, (5) the item was likely to discriminate by severity of condition, (6) the item was likely to discriminate among known population groups, (7) the item was frequently mentioned by participants, and (8) the item was relevant to everyone with the condition. This resulted in 125 items that were retained and presented to the four adolescents with CFDs who participated in the cognitive debriefing exercise, a group of parents, and the professional panel for assessment. Each of these individuals selected the 30 items that they thought best captured the QoL issues for adolescents with CFDs.

This process resulted in 71 perceptual items (across seven categories). The items in the surgery category comprised the Youth Quality of Life Instrument–Craniofacial Surgery (YQOL-CS) module ($n = 32$ items), whereas those in the remaining six categories comprised the Youth Quality of Life Instrument–Facial Differences (YQOL-FD) module ($n = 39$ items). The surgery items were divided into those relating to past surgery ($n = 20$) and future surgery ($n = 12$). The core group of investigators selected the YQOL-FD perceptual items for which verifiable items could be written to comprise a contextual set of 18 items. Also, two contextual YQOL-CS items were written to capture expectations for having future surgery and the delay of surgery in the past.

Tables 1 and 2 show the module domains and example perceptual items from each domain. These items are on an 11-point response scale with adjectival anchors of “not at all” (0) to “a great deal” or “completely” (10).

A readability analysis was conducted of the YQOL-FD and YQOL-CS modules using the Homan-Hewitt Readability Formula (Homan et al., 1994). The Homan-Hewitt Formula is designed especially for assessing single-sentence items. The grade reading levels of the modules are as follows (± 0.5): YQOL-FD contextual = 4.6, YQOL-FD perceptual = 3.4, and YQOL-CS perceptual = 4.1.

DISCUSSION

In this study, adolescents with CFDs proved to be interested and able to discuss how their lives are affected by having a CFD and resulted in the development of two condition-specific QoL modules: the YQL-FD module and the Craniofacial Surgery module. The facial differences module includes positive aspects that youth with CFDs consider important to their QoL (e.g., acceptance of others, empathy) and also providing information on the areas of their life that have a negative effect

TABLE 1 Youth Quality of Life Instrument—Facial Differences Module: Domains and Example Perceptual Items*

<i>Domain</i>	<i>Example Perceptual Items</i>
Coping	"I am a tougher person because of my facial difference." "Because of my facial difference, I need to prove myself to others."
Stigma and isolation	"Because of my facial difference, people have a hard time seeing the real me." "Because of my facial difference, I feel left out of things."
Intimacy and trust	"My facial difference has made it difficult to find really good friends." "Once people get to know me, they forget I have a facial difference."
Positive consequences	"My facial difference has helped me to accept others for who they are." "Because of my facial difference, I am in tune with other people's feelings."
Self-image	"I dislike looking at pictures of myself because of my facial difference." "I feel attractive to others, even though I have a facial difference."
Negative emotions	"Because of my facial difference, I feel I have no control over my life." "Because of my facial difference, I have more anger inside me than most people know."

* Scale = "not at all" (0) to "a great deal" or "completely" (10).

(e.g., stigma, isolation). The craniofacial surgery module highlights both attitudes toward surgery and expectations of how surgery might affect their QoL. These instruments, once validated, may provide useful information in the evaluation of surgical and psychosocial interventions.

Currently it is difficult to build on the successes of patients and families because in the past we have tracked primarily pathology and maladjustment. Adolescent interviewees in this study reported three positive aspects related to having a CFD: learning to cope with having a CFD and becoming a tougher person as a result, learning to be more accepting of others, and becoming more in touch with other people's feelings. Learning to cope relates to the concept of resilience, which figures prominently in the contemporary adolescent research literature (Olsson et al., 2003) and has been described simply as "normal development under difficult conditions" (Fonagy et al., 1994, p. 233). The positive aspects of having a CFD that were discussed by these youth correspond with findings from studies of adults with CFDs (Eiserman, 2001; Kelton, 2001). Thus, both internal and social dimensions were found to be affected positively for some of the adolescents with CFDs. Although this was not true for all, as further evidenced by findings in the companion article by Topolski et al. (2004) concerning sense of self, we believe that including these issues gives a greater amount of balance to assessment.

An important issue to consider with regard to these modules is how and in what contexts they are best used. After being psychometrically validated, we believe they will have much utility in clinical trials for determining the effect of surgical and psychosocial interventions on QoL. We believe they will also have potential use in day-to-day clinical practice as a com-

TABLE 2 Youth Quality of Life Instrument—Craniofacial Surgery Module; Example Perceptual Items*

"I get angry at my parents for making me go through craniofacial surgery"
"I feel my craniofacial surgeries have been a waste of time"
"After I have my craniofacial surgery I won't have to worry about being teased"
"My craniofacial surgery has improved my life"
"I take part in making decisions regarding my craniofacial surgery"
"I feel I need additional craniofacial surgery in the future"

* Scale: "not at all" (0) to "a great deal" or "completely" (10).

plement/supplement to deficit-oriented measures and in the assessment of "surgery readiness" from a psychological perspective. It is important, however, that the modules are not used in lieu of in-depth psychosocial assessment, support, and intervention in the clinical setting. With regard to clinical trials, it is important to avoid misuse of results by insurance companies and medical equipment corporations. We need to be prepared for the ramifications if in a clinical trial the QoL-related outcomes are not what we thought they would be.

An important contribution of this study is the conceptual and methodological separation of adolescents' views of craniofacial surgery, specifically from their assessment of how CFDs have affected their lives more generally. Youth's voices are often translated through complex, intimate interactions among parents, surgeons, and other clinicians, and there is often an assumption made by parents and clinicians that surgery will lead to an improvement in QoL. The patient, however, may be dissatisfied with the surgical result but still experiencing improvement in his or her life that may or may not be attributable to the surgical change. Surgery-related QoL as assessed with this module includes issues of the actual experience of having surgery and how it affects satisfaction. Patient concerns about such issues as missed life opportunities or pain will understandably affect the overall assessment, but these issues have rarely been examined.

There are at least two limitations of this study. First, there is the potential for an ascertainment bias in the adolescent participants who were recruited into the study. As noted above, youth with CFDs have been observed to be more shy and reticent than their peers who are not affected, and those who volunteered to participate in the study and tell us about their lives and experiences may be unrepresentative of their less outgoing peers with CFDs. A second potential limitation of this study is that the replicability of the conceptual domains has yet to be established.

In conclusion, using grounded theory and established qualitative methodologies, two QoL measures specific to adolescents with CFDs were developed and are ready for validation in a second phase of this outcomes research development process. Validation refers to testing the adequacy of the measures' psychometric properties, including scale structure; test-retest reliability; discriminant and convergent validity; responsiveness to change; cultural adaptation (Mexican, Spanish, and British English); and respondent burden. The validation study, also funded by the National Institute of Dental and Craniofacial Research, is currently underway at five sites (four in the

United States and one in the United Kingdom), scheduled for completion in spring 2005. After the psychometric properties of the modules have been established, the next step will be to take them into clinical trials to test the effectiveness of surgical and psychosocial interventions in enhancing QoL.

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