Critical Issues in Craniofacial Care: Quality of Life, Costs of Care, and Implications of Prenatal Diagnosis

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Since the 2000 Surgeon General's Report on Oral Health (SGROH), substantial areas of inquiry relative to individuals, especially children and youth, with orofacial clefts and other craniofacial conditions have emerged. These areas include access to and cost of care, stigmatization and quality of life, and social and ethical issues around prenatal diagnosis. This update on the 2000 SGROH examines what we have learned about the cost and ability to access cleft and craniofacial care, prenatal diagnosis, and how quality of life is impacted by these conditions and the burden of care. The development of new research tools to assess quality of life since 2000 will permit further study of the impact of oral and craniofacial conditions on children and families and the effect of treatment on quality of life. Despite a better understanding of the higher use of services and increased costs of care for families of children with craniofacial conditions, major gaps in research must be addressed to assist with program planning and policy development for these groups of children and their families. Further work is also needed to assess the cost-effectiveness of craniofacial team care and to better understand family experience with accessing needed care. Finally, prenatal detection and diagnosis of clefts and craniofacial conditions have advanced dramatically, and the roles of craniofacial professionals and teams have been affected. New understandings of prenatal diagnosis and genomic sciences are redefining genetic counseling, therapy, and future preventive initiatives.

KEY WORDS: access to care; cost of care; craniofacial conditions; orofacial clefts; prenatal diagnosis; quality of life

Academic Pediatrics 2009;9:427–32

The 2000 Surgeon General's Report on Oral Health (SGROH) identified profound disparities in oral health and access to care for vulnerable populations, including individuals with disabilities and other special health care conditions, and the need for more information about these populations.[1] The SGROH also highlighted the impact of oral and craniofacial conditions (CFC) on overall health and quality of life (QOL) and called for more understanding of these relationships. Since then, studies have advanced our understanding in several key areas, including measurement of QOL in children and adolescents with oral and CFC and the costs of care for these children. Additionally, advances in technology in the last decade have made prenatal diagnosis of CFC increasingly common, raising issues with profound ethical and social implications for all involved in their care. Although notable advances also have been made in other areas, including the basic sciences, here, we focus on the social, economic, and ethical issues.

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Received for publication May 13, 2009; accepted September 22, 2009.

Orofacial Clefts and Other Craniofacial Conditions

Orofacial clefts (OFC) are the most common of the CFC and are the best studied. They include cleft palate and cleft lip with or without cleft palate and are among the most common birth defects in the United States.[2] OFC can affect physical growth and development of teeth, speech, hearing, feeding capabilities, and psychomotor and cognitive skills, creating both physical and psychosocial challenges and significant costs for these children and their families. Moreover, these concerns change over time as children develop, complicating long-term outcome studies and making costs of care difficult to assess over the life span.[3–10] The complex, interrelated health and psychosocial issues that arise highlight the need for coordinated, interdisciplinary team care, as recommended by a previous Surgeon General's Report on Children with Special Health Care Needs[11] and by other standards in the craniofacial field.[12] Such care is difficult to ensure in a health care system that incentivizes short-term, acute interventions over long-term team management aimed at optimal future health and QOL and functioning in society. Increasingly, a better understanding of QOL and more accurate data on health care use, access to care, and costs of care are being examined to help plan for these children's care.

QUALITY OF LIFE

Over the past decade, the construct of QOL has been explored in the context of CFC. Considered part of overall health, QOL can be defined as individuals' "perceptions of

their position in life in the context of the culture and values in which they live, and in relation to their goals, expectations, standards and concerns."[13,14] Recent studies of QOL among adolescents with CFC have both furthered understanding of factors mediating their experience and resulted in new measurement tools. The Youth Quality of Life Instrument, Research Version (YQOL-R) was developed to reflect a broad social understanding of QOL in youth and is a multidimensional instrument that assesses cultural, social, physical and psychological well-being.[15] The Youth Quality of Life—Facial Differences (YQOL-FD) module specifically examines perceptions about the impact of facial differences on QOL.[16] Developed through focus groups and detailed patient interviews, this patient-centered research revealed adolescents' highly charged emotions, both negative and positive, related to their conditions, to the experience of undergoing repeated surgeries to "look better," and to their often being excluded from the surgical decision-making process.[17] Studies that use these newly developed instruments can further our understanding of how specific treatments impact QOL and will clarify the burden of care; these new studies will also open the possibility for cross-site or multination comparisons.

Stigma, Appearence, and Resilience

Research on stigmatization, facial appearance, and psychological resilience has heightened our awareness of the subjective experiences of individuals with CFC. Because the face is central in interaction and interpersonal perception,[18–21] facial differences that accompany CFC may be particularly likely to elicit stigma,[22] thus influencing QOL. In one study that used the YQOL-R, facial difference correlated to lower scores on the measure, relating to concerns such as being left out and feeling unwelcome by peers, less important, and less safe at school.[23] Appearance influences interactions and relationships,[24–26] and several studies show that the type of OFC or CFC may relate to judgments of attractiveness[27–29] and stigmatization.[30] The pressures for conformity to a common societal standard of appearance or function are evident in advertising and media images and in the social pressures placed on persons with disabilities to normalize by surgical and other treatment efforts.[31] Concerns over how these pressures drive care to "surgically shape children" prompted the creation of an in-depth working group to consider these issues at the Hastings Center, a bioethics research institute.[32]

Awareness of stigma and QOL in persons with OFC and CFC has also led to new craniofacial social science models to understand resilience and the development of healthy identity. Historically, much craniofacial research has focused on deficits, limitations, handicaps, and challenges.[33] Although such studies clarified the biological and psychosocial challenges that children with CFC confront, the emerging focus on resilience, strengths, and optimism are moving some researchers to understand how to maximize human potential in persons with OFC.[32,33] Social scientists have begun to ask new questions that probe the sociocultural sources of resilience, including how family life, culture, myth, education, courage, faith, and/or humor arm children to succeed in adult life.

To address stigma in the social arena, educational interventions in schools have sought to alter social attitudes about appearance.[34] Nonprofit advocacy groups have developed programs such as AboutFace for classroom teaching about CFC and the impact of "looking different."[35] This group and others, including the Cleft Palate Foundation, also offer information about CFC to parents, professionals, and affected persons.[36,37] The Cleft Palate Foundation also uses television advertising to portray persons with cleft lip and palate in successful careers and offer affected individuals positive role models. Such efforts seek to reduce the stigma experience of CFC and demonstrate that altered appearance is not necessarily a permanent impediment.

Oral Health and Quality of Life

Since the 2000 SGROH, research tools for the general population of children that measure the impact of oral health on a child's health and QOL have been developed. The Child Oral Health Impact Profile is the first validated instrument to incorporate both negative and positive aspects of health in measuring the impact of oral conditions in children aged 8–17 and their caregivers.[38–41] Oral health impacts on QOL had been studied in adults,[42,43] and the Child Oral Health Impact Profile allows for the study of oral health conditions in children. Although the value of self-report outcome measures is clear and central to a more expanded construct of QOL as previously described, reports from parents or caregivers may be the only way to learn about QOL in younger children who are unable to understand or provide such input.

Continued research is needed to better understand the contribution of oral health to overall health, including QOL, the resultant human costs of disorders occurring in the craniofacial complex, and to help elucidate the best approaches to care and assessment of outcomes.

ACCESS TO AND COST OF CARE

Quality of life is one important outcome measure to assess the impact of CFC and OFC on children and families. Other measures include access to and cost of care. Children with special health care needs, of which children with CFC represent a subset,[44] have greater health service use, incur greater costs, and experience more barriers accessing care than children without such needs.[45–47] However, national data on health service use and costs that pertain to children with special health care needs and selected subgroups of this population are limited and do not specifically address children with OFC and CFC.[45,48–50] In addition, few data are available that describe variability in service use and cost by child characteristics, such as age and diagnosis for children with CFC and OFC.

Cost Studies on Children with OFC

Before 2000, only 2 studies had been conducted on health service use and costs of children with birth defects,