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,

including OFC.[51–53] One study determined costs associated with medical, mortality, morbidity, disability, and developmental services.[51,52] Even though they are considered the most comprehensive studies of cost of birth defects, these dated reports did not provide details on cleft type, associated syndromes, or other CFC.

Since the SGROH, several studies examined health care service use and provided more updated costs of care for children with OFC. Two studies stratified costs by cleft types and calculated the number of services received over the first few years of life. Limitations of these studies included small sample size, selection bias, and examination of charges rather than the amount for services rendered.[54,55]

Four recent studies measured hospitalizations and medical costs of children with OFC. Two studies used 2003 and 2004 data from the Healthcare Cost and Utilization Project to examine hospitalizations and costs in children with OFC in the United States. One of these studies considered a variety of ages,[56] and the other examined costs in the newborn period.[57] Two studies examined expenditures from a payer perspective for children with OFC and compared the results to unaffected children, and stratified the results by 3 cleft categories and presence of other anomalies.[58,59]

Timeliness of and Referral to Services and Barriers to Care

Services and treatment for children with OFC and other CFC can vary depending on severity, presence of associated syndromes and/or other birth defects, and child's age and other needs.[4] Recommended guidelines for services and treatment for children with these conditions have been set forth by the American Cleft Palate-Craniofacial Association and others.[4,12,60,61] To date, only 2 studies have examined the timeliness of such services in accordance with the American Cleft Palate-Craniofacial Association recommendations.[62,63] Referral of children with CFC can affect receipt of services and thus costs of care. Two recent studies examined referral to specialized services among children with OFC.[64,65]

Many financial and nonfinancial factors can impede health service use and receipt of timely services among children with OFC and CFC, but data on perceived barriers to care are lacking. Only 2 qualitative studies have examined access to care specifically among children with birth defects, which included OFC, yet both focused on dental care.[66,67] To date, no study has specifically addressed barriers to health care among children with CFC.

In summary, since the SGROH, we have a better understanding of the extent to which children with OFC utilize more health services and have greater health care costs than children without this condition. We also have updated cost and expenditure information on a variety of age groups, a range of health service categories, and privately and publicly insured children. We have improved on these estimates by stratifying by 3 cleft types and by presence of other anomalies.

However, gaps remain in our knowledge of the effect of payer status and other maternal, child, and system factors on health service use and cost for these children. Four areas need special study. Service use and cost of care beyond the neonatal period and over the life span should be studied to better understand the financial burden of CFC. Additional information on indirect and other costs, such as caregiver costs and out-of-pocket expenses, and costs of early intervention services, is needed. Further understanding of the effectiveness of team care and care coordination provided by craniofacial teams and centers, medical homes, and ancillary services for individuals with CFC is essential. Finally, additional further research on the identification and referral of children with CFC is warranted to improve the timeliness of services and QOL and health outcomes in these children.[63,65] These gaps in the literature were recently identified by the Centers for Disease Control and Prevention (CDC) as research priorities for children with OFC and CFC.[68] Improving our knowledge in these areas can help us better assess the cost-effectiveness of treatments and aid in policy development to improve the quality and access to health care for patients and families with CFC.[63] Understanding patterns of health service use, costs, and expenditures can also help to target populations in need of services and appropriately allocate health care resources.

The high costs of caring for children with CFC or other birth defects can also raise issues about the intrinsic value of these children's lives, and society's and families' commitment to provide for this care. Nowhere are these questions more apparent than when a family receives a prenatal diagnosis of a CFC or other congenital abnormality.

PRENATAL DIAGNOSIS: ETHICAL ISSUES FOR FAMILIES, TEAMS, AND SOCIETY

With the increasing sophistication and availability of prenatal diagnostic technologies, such as transvaginal ultrasound, chorionic villus sampling, amniocentesis, and alfa-fetoprotein testing, more genetic and congenital conditions are being detected during pregnancy. On the basis of prenatal diagnostic findings, parents may choose either to continue the pregnancy with a new awareness and armed with helpful information, or to terminate the pregnancy. However, little is known about how families and society deal with the inevitable ambiguity, uncertainty, and difficult decision making that occurs with a prenatal diagnosis.[69–71] Indeed, it is not known whether prior knowledge of a craniofacial condition is an advantage to families.[72] Knowing in advance about a future child's special needs allows a parent to anticipate future social pressures regarding the child's integration and ultimate QOL. If parents perceive that they or the child will be greeted with acceptance, they may feel supported. If they perceive that they or the child will face a high level of stigmatization and prejudice, they may feel isolated and marginalized.[73]

As prenatal diagnosis becomes more accurate and available at earlier times in the pregnancy, other issues will arise, such as who will pay for prenatal diagnosis and who will have access to it. Craniofacial centers have

reported ethical issues related to prenatal diagnosis and nonlethal birth defects.[74–76] Fundamental questions about the rationale for prenatal diagnosis have been raised. For example, can a repairable condition such as cleft lip be seen as the basis for the termination of pregnancy? Is prenatal diagnosis meant to detect seriously impairing or life-threatening conditions, or is it meant to identify genetic or congenital traits that are compatible with a high-quality life span? Where the line lies between these 2 groups is culturally and socially determined.[77] Is prenatal diagnosis a vehicle to normalize human difference and reduce variability in the human genome, or is there inherent, but unappreciated, value in the variation expressed in the human genome? Is it possible to identify genetic differences between people, but not to seek to normalize non-life-threatening imperfections?[78]

For craniofacial team professionals who focus on delivering care to individuals with CFC, the ethical issues can be acute. There are different opinions about the most appropriate relationship between the prenatal diagnostic advisor and the treatment team. Some craniofacial professionals hold that team professionals should focus on promoting the QOL for all their patients and not become engaged in advising parents about prenatal diagnosis and about the possible termination of pregnancy. Others say craniofacial team members have unique knowledge and information that parents will need to guide their decisions about whether to bear and how to treat the affected child.

For the affected individual, the discussion about the value of a life lived with a cleft raises the fear that somehow their life was not worth it, that somehow they have placed a burden on their parents that was just too heavy. As Eiserman and Strauss[76] suggested, "perhaps, some affected persons wonder 'what would my family's life have been like if they had been spared all the expense and heartache that I brought with me?'"

The manner in which professionals present information regarding a prenatal diagnosis may determine how parents will respond. Ideally, prospective parents should receive nonjudgmental and unbiased information, support, and advice from health professionals who are trained to counsel and support decisions.[75,79]

Craniofacial team professionals can help parents of unborn children evaluate fetus's condition and project the range of possibilities that might occur in their child's life. They can help guide parents in their search for information, assure their data are accurate, and interpret their findings. Complex prenatal diagnostic data may provide too much information for families to handle on their own.

The craniofacial team's ability to manage prenatal diagnostic and future genome technologies will depend on their insight and on their vision of what constitutes a "good" life. Recent awareness of the impact of prenatal diagnosis for OFC and CFC has raised interest in themes that relate to QOL as well as social and ethical issues. In the past decade, craniofacial centers and teams roles have been redefined by the emergence of prenatal diagnostic technologies and will continue to be altered with the advancement of such technologies.

RECOMMENDATIONS AND CONCLUSIONS

Since the 2000 SGROH, research relating to CFC has advanced, and new understandings have emerged regarding health services research, QOL, stigmatization, and the impact of prenatal diagnosis. These new research areas have brought new investigators to apply their skills and methods to craniofacial research. The involvement of health services researchers, epidemiologists, social scientists, geneticists, and ethicists has created opportunities to enhance QOL, team care, and health outcomes for children with CFC.

Important care dilemmas have driven the research and discussions related to QOL, prenatal decisions, costs of care, and access to care. Health care use, timeliness of services, costs, barriers to care, QOL, outcomes, and prenatal diagnosis of OFC were recently identified as research priority areas by the National Center on Birth Defects and Developmental Disabilities at the CDC.[68] Furthermore, better estimates of costs and expenditures, including the effect of private and public payers on outcomes and caregiver costs and out-of-pocket expenses, for these children and their families are important for developing policies and programs and for enhancing health and QOL. We recommend also employing techniques, such as geographic information systems analysis and geographical variables, to determine any regional differences in access to care. We also suggest examining maternal, child, and system factors that may contribute to barriers to care and timeliness of services. Such information can lead to targeting populations experiencing unmet service needs and improving service availability and delivery.

We recommend further exploration of QOL outcomes among school-aged children with CFC, as well as ethical, societal, and policy implications of prenatal diagnosis. We also recommend building stronger relationships with state birth defects registries and craniofacial teams to refer children with CFC to services. Promoting and establishing referral systems to craniofacial teams and conducting craniofacial health surveillance among children with these conditions are part of Healthy People 2010 objectives 21-15 and 21-16.[80]

ACKNOWLEDGMENTS

We thank Dr Wendy E. Mouradian for her insightful comments and careful manuscript reviews and Vanessa White for her valuable literature review assistance. Dr Cassell thanks the National Center on Birth Defects and Developmental Disabilities at the CDC for providing funding for the studies on health service use and expenditures conducted in North Carolina (grant U50/CCU422096).

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