

‘Burden of Oro facial clefts, Quality of life and Costs- a review article’.

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ABSTRACT

BACKGROUND

Oro facial clefts (OFC) are the most frequent birth defects worldwide. Cleft lip with/or palate are the major human oro facial congenital malformations seen in live birth as well as in still birth. It affects about 1.5 per 1000 live births (250,000 new cases per year) worldwide, with tremendous variations across geographic areas and ethnic groups.

Cleft lip and palate represent a major public health problem due to the possible associated life-long morbidity, complex etiology and the extensive multidisciplinary commitment required for intervention. Affected persons have a range of functional as well as aesthetic problems. These include feeding difficulties at birth due to problems with oral seal, swallowing and nasal regurgitation, hearing difficulties due to abnormalities in the palatal musculature, and speech difficulties due to nasal escape and articulation problems.

These common birth defects that may impose a large burden on the health, quality of life, and socioeconomic well-being of affected individuals and families. They also result in significant healthcare use and costs. However, OFC impose a large psychosocial and economic burden on affected families and society and associated with several health problems and complications.

BODY

In Bangladesh few data are available to identify determinants of variability in service use and costs, including patient (cleft severity, presence of other conditions), family (socioeconomic and demographic), and area-level (healthcare availability and access) characteristics.

There are approximately 300,000 cleft lip and palate (CLAP) sufferers in Bangladesh amongst a population of 120 million. These common birth defects that may impose a large burden on the health, quality of life, and socioeconomic well-being of affected individuals and families. They also result in significant healthcare use and costs. However, OFC impose a large psychosocial and economic burden on affected families and society and associated with several health problems and complications.

CONCLUSION

In Bangladesh few data are available to identify the determinants of variability in service use and costs, including patient (cleft severity, presence of other conditions), family (socioeconomic and demographic), and area-level (healthcare availability and access) characteristics. Understanding patterns of health service use and expenditures can help to target populations in need of services, assess the cost-effectiveness of treatments, and develop policies to improve the cost-effectiveness of and access to healthcare for patients and families with OFC.

KEY WORDS

Oro facial clefts, Quality of life, Economic burden, Oral Health Related Quality of Life.

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I. BACKGROUND

Oro facial clefts (OFC) are the most frequent birth defects worldwide. Cleft lip with/or palate are the major human orofacial congenital malformations seen in live birth as well as in still birth. Cleft lip and palate represent a major public health problem due to the possible associated life-long morbidity, complex etiology and the extensive multidisciplinary commitment required for intervention. The term "orofacial cleft" (OFC) covers a wide range of disorders affecting the lips and oral cavity which manifest in the new born infant. OFC involves structures around the oral cavity which can extend on to the facial structures resulting in oral, facial and craniofacial deformity. The main categories are isolated cleft palate (CP) and cleft lip with or without cleft

palate (CL/P). Additionally, OFCs may be left- or right-sided (unilateral), or both sided (bilateral). Both types may present as part of a syndrome or other associated abnormalities.

Oro facial clefts are common birth defects that may impose a large burden on the health, quality of life (including the general and mental health of the affected individuals and caregivers), and socioeconomic well-being of affected individuals and families. They also result in significant healthcare use and costs. Affected person have a range of functional as well as aesthetic problems. These include feeding difficulties at birth due to problems with oral seal, swallowing and nasal regurgitation, hearing difficulties due to abnormalities in the palatal musculature, and speech difficulties due to nasal escape and articulation problems. These cleft defects can be surgically repaired in childhood, but residual deformity due to scarring and abnormal facial development results in continuing functional and psychosocial problems. Thus, clefts have a prolonged, adverse influence on the health and social integration of affected individuals.

It affects about 1.5 per 1000 live births (250,000 new cases per year) worldwide, with tremendous variations across geographic areas and ethnic groups.

Prevalence rates vary as a function of ethnic, geographical, and socio-economic factors, and mothers' general health status. A complex etiology of genetic and environmental factors likely contributes to these problems.

Oral clefts considered a debilitating condition that are associated with difficulties in feeding, growth, cognitive development, speech, hearing and behavioral impairments. The needs for the wide surgical, dental, speech, social, and medical involvement emphasize the importance of understanding the underlying determinants of these defects to allow optimizing the treatment options and predicting the long-term course of the affected individuals development. These may significantly increase the risk of neonatal and infant mortality, especially when present with other birth defects. Furthermore, oral clefts may increase long-term all-cause mortality and suicide risks.

Cleft lip and palate occurs in about 1 to 2 per 1000 births in the developed world². Birth prevalence of CL/P and CP) in India is 1.10 per 1000 and 26,950 per year. Cleft lip, with or without cleft palate, affects one in 700 babies annually, and is the fourth most common birth defect in the U.S. Clefts occur more often in children of Asian, Latino, or Native American descent. Compared with girls, twice as many boys have a cleft lip, both with and without a cleft palate. However, compared with boys, twice as many girls have cleft palate without a cleft lip.

Cleft Lip/Palate is very prevalent in Bangladesh. Each year 5,000 children are born with cleft in Bangladesh- Smile Train. Whereas in most countries it is solely a genetic issue, in Bangladesh it is most often a result of malnutrition of the mother while she is pregnant. When a baby is born with cleft, the mother is blamed as being cursed or of having done something wrong while pregnant to cause her baby's deformity. Most parents are unaware that the deformity is even treatable. Most of these unfortunate children live their lives in shame – unable to eat properly, talk, go to school and are taunted by many. They also impose a large psychological and economic burden on families and society. Many children born with cleft are abandoned at birth by their parents.

There are approximately 300,000 cleft lip and palate (CLAP) sufferers in Bangladesh amongst a population of 120 million. The vast majority of these patients cannot afford and do not have access to even basic surgical repairs or cleft related services.

Cleft lip and palate may be perceived to be a life threatening abnormality and there may be little awareness of the fact that clefts can be surgically repaired with considerable success both aesthetically and functionally.

Several studies have found reductions in the quality of life and psychosocial performance among affected individuals that is partly related to low satisfaction with facial appearance.^{22, 23, 24, 25,26, 27, 28, 29} The effects of oral clefts may also extend through adulthood and reduce psychosocial, educational, and economic achievement.

These common birth defects that may impose a large burden on the health, quality of life, and socioeconomic well-being of affected individuals and families. They also result in significant healthcare use and costs. However, OFC impose a large psychosocial and economic burden on affected families and society and associated with several health problems and complications. Besides that throughout history and around the world, people with disabilities have been subjected to cruel and inhumane treatment.

Oral Health Related Quality of Life (OHRQoL) is an extension of HRQoL that more specifically measures the impact of oral disease on the person's physical and social functioning. OHRQoL is an integral part of general health and well-being. In fact, it is recognized by the World Health Organization (WHO) as an important segment of the Global Oral Health Program (2003). Health-related quality of life (HRQoL) is a patient-reported outcome that helps clinicians assess how patients are affected by their illness or condition by quantifying functional and psychosocial well-being of the patient. Measuring HRQoL is becoming an increasingly important mechanism for evaluating different aspects of disease management.

Though data and information on these birth defects including the assessment of their quality of life and economy related impacts have been conducted in different countries but these type of information are not

available at the national level of Bangladesh. A number of studies have been published reporting the frequency of birth defects in defined populations and geographic areas.

The primary goal of this paper is to review and discuss the health-related quality of life (HRQL) of the cleft lip and palate patients and their caregivers, socioeconomic outcomes, health outcomes, and perceived barrier to the healthcare service utilization and costs. A secondary objective is to identify primary research gaps and potential study designs to address these gaps.

BODY:

Cleft lip and palate are considered one of the most common birth defects that possess significant medical, psychological, social, and financial implications on the affected individuals and families. Clefts have a complex etiology with both genetics and environment playing a role. Risk factors such as folic acid deficiency, maternal age, and maternal smoking have been linked to the development of clefts. In addition to the aesthetic disfigurement, a child with cleft lip and/or palate suffers substantial functional morbidity such as restricted maxillofacial growth, speech anomalies, swallowing and feeding difficulties, hearing loss and/or recurrent ear infections. Although not generally life-threatening, living with a cleft elicits a significant health burden.^{3,4} Cleft defects have a long term, adverse influence on the health and social integration of affected individuals because even though they can be surgically repaired early in childhood, residual deformity due to scarring and abnormal facial development results in continuing functional and psychosocial problems.^{4,5}

In many high-income countries (HICs), active surveillance systems are in place, and several CLP studies have been conducted that provide epidemiological trends and prevalence estimate.^{6,8-11} Several studies have reported on access and utilization of treatment and health care services for CLP¹²⁻¹⁶; standards and quality of care and long term health outcomes¹⁷; the clinical profile of cases, and the composition and interaction among healthcare team members in the treatment of CLP.¹⁸⁻²¹

Presently there is an emerging body of literature on CLP in low-and-middle-income countries (LMICs), focusing on the epidemiology of CLP²²⁻²⁹, quality of life, treatment and care of individuals with CLP, health care access, service challenges, and resource constraints.³⁰⁻³³

The treatment of an individual with cleft lip and/or palate is a very difficult task and most of the time requires close long-term collaboration among various specialists. The multidisciplinary teamwork may eventually lead to a successful treatment outcome with a minimum of procedures and optimal cost-effectiveness.³⁴ However, it is quite usual that lack of long-term treatment planning from birth to adulthood and standardized surgical protocols result in poor esthetic and functional treatment outcomes.³⁵⁻³⁷ The presence of an orofacial cleft significantly decreases overall ohrql, functional well-being, and social-emotional well-being in children and adolescents, with similar impact in patients and parents. The purpose of this review is, these studies may provide important preliminary insights into the relationship between OFC and HRQL.

Variations in the HRQL preferences of patients, parents and health professionals were reported in the previous studies. A study by Berket al. (2001) found that compared to siblings without OFC, adults with OFC may be more disadvantaged with respect to social affiliation and adaptation.³⁷

Marcusson investigated a Swedish sample of 68 adults with OFC and 66 adults without OFC. Compared to the gender- and aged-matched control group, adults with OFC had significantly lower scores for quality of life, family life, private economy, global life, disturbance to life, well-being, and social contacts. However, more practical and tangible aspects of the daily lives of the adults with OFC were not affected. He concluded that adults with OFC reported a fairly good life adjustment in spite of the presence of the facial malformation.³⁸

Danino et al. investigated 82 French adults with repaired CLP and found that, compared to adults without CLP, people with CLP did marry later, displayed a delay in scholarship, had a lower income, and reported a significant delay in their independence process from their parents.³⁹ These findings do match those based on observations of a Norwegian sample of 233 adults aged 20 to 35 with repaired OFC: Compared to the general population, adults with OFC were less likely to marry and, if they did so, to marry later in life, giving birth to fewer children. (Ramstad et al. 1995a).

Some studies found decreased quality of life of adolescents with congenital and acquired facial malformations compared with unaffected adolescents as well as frequent reporting of stigmatization experiences. Quality of life decreased with the individuals' perceptions of increasing severity of facial malformations.⁴⁰⁻⁴⁵

Sinkoet al at 2005 found lower HRQL among a sample of Chinese adults with repaired CLP aged 18–30 years, who wanted to receive more treatment compared with those who did not. Further, the study found that CLP impacted emotional and social functioning. Marcusson et al evaluated the HRQL in a sample of adults with CLP and unaffected adults from Sweden.³⁸ The authors reported significantly lower HRQL in the affected

sample compared with the unaffected sample. The study found higher HRQL among adults who were more satisfied with their facial appearance.

A higher risk of hospital admission because of mental health complications among adults with CP and CL/P were found compared with unaffected adults. These studies strongly suggest that OFC imposes a large burden on the health of affected individuals throughout the life span.⁴¹

Oosterkamp et al. evaluated the HRQL of a small sample of affected adults with bilateral CLP and unaffected adults from the Netherlands and found no significant differences in HRQL scores between the two groups, but higher HRQL among adults who were satisfied with their appearance.⁴²

Differences in the HRQL preferences of patients with craniofacial conditions and their parents have also been reported.⁴³

Several studies have reported psychological challenges among children, adolescents and young adults with OFC. Speech problems and concerns about esthetics are thought to contribute to these challenges. Some studies have also found increased social anxiety among affected adults, although findings have varied between studies (reported a higher mortality rate as a result of suicide among individuals with OFC in Denmark compared with unaffected individuals).⁴⁴

Several cost perspectives exist, including the healthcare system, which includes direct costs, and societal perspective, which includes all costs, for example, caregiver costs and out-of-pocket expenses. Another viewpoint is the payer perspective, such as public and private health insurance, which measures costs with payments to providers. Depending on which perspective one is using, costs, charges, or expenditures are the appropriate measure of effect.

Because costs and expenditures are often used as a proxy for healthcare service utilization and are usually the most salient issues in terms of service delivery, program planning and policy development, it is necessary to focus primarily on costs and expenditures of patients with OFC and health service utilization. Differences in the results of these studies are attributable to using different: payers; definitions for healthcare service categories; ages; and units of analysis, such as hospital discharge (hospital stay) compared with an individual.

Brand et al. in 2009 showed that children and adolescents with CLP were six times more likely to report difficulties in interactional competencies compared to controls without CLP. Stigma experiences, social problems, symptoms of anxiety and depression, dissatisfaction with facial appearance, withdrawal and internalizing problems (Pope & Snyder, 2005), and shyness have also been reported.⁴⁵

In a cross-sectional study, Hunt et al. (2006) assessed the psychological functioning of patients with CLP, comparing 160 children and young adults with an age- and gender-matched control group. The main results were that, compared to controls without CLP, children and young adults with CLP reported greater behavioral problems, increased symptoms of depression, and a lower satisfaction with facial appearance and speech. Moreover, these participants indicated that they were more likely to be teased in social settings. No significant differences between participants with and without CLP were found for anxiety and self-esteem. However, independently of the presence of CLP, having been teased predicted significantly poorer psychological functioning. Hunt et al. (2006) concluded that, as part of routine cleft care, children and young adults with CLP require psychological assessment, specifically focusing on their experiences being teased.

Feragen in 2009 reported that cleft types were related to the frequency of additional physical and psychological difficulties, pointing to a need to differentiate between types of OFC. They were also able to show that psychosocial resilience was related to adequate emotional functioning, high satisfaction with appearance, and low reported frequency of being teased. Last, compared to children and adolescents with other than OFC, those with OFC registered higher scores for social problems and deficits in social and academic competencies. Moreover, adjustment seemed to be related to appearance and speech deficits, which in turn appeared to be associated with poor peer relationships. Specifically, poor adjustment was observed in those children reporting increased teasing by peers.⁴⁴

In Bangladesh few data are available to identify determinants of variability in service use and costs, including patient (cleft severity, presence of other conditions), family (socioeconomic and demographic), and area-level (healthcare availability and access) characteristics. Unfortunately, there is a stark contrast between high- and low-resource areas with barriers to treatment including cultural beliefs, geography, and financial support.

Social stigma is context dependent and is based on cultural perception of the cleft. Blame of the parent or the perception that the cleft is from a punishing super-natural force negatively influences the perception of the child with CLP. A child with CLP is subjected to bullying, rejection, and social isolation, even at times, from their family. This torment extends to school leading to a dropout from education and adds a further barrier to reaching their ideal employment. The inability to fully participate in the workforce translates into a substantial economic burden and lost productivity. Psychological distress naturally follows, with children being more

affected than adults. Depending on the cultural context, the impact of cleft disease may or may not have an impact on marital status.⁴⁵

In some previous studies it has been found that etiology for cleft is seen to be punishment on the parent, blame on the parent, or personal conduct of the parent including the parent slipping and falling or force to the fetal face. One belief was that the father cut open the mouth of a fish to remove a fish hook whereas the mother was pregnant. Other causes include the effect of "God's will," supernatural forces, evil spirits or ancestral spirit exposure to an eclipse, black magic, or a contagion. Further, children with CLP may not be seen as human. When asking the general community about cleft, very few knew that it affected the lip and face, much less its etiology.⁴⁶⁻⁴⁹

Sheikh Hasina National Institute of Burns and Plastic Surgery are working with Bangladeshi specialist to create a sustainable center to treat children with cleft lip and palate. They have funded the appointment of the first cleft speech and language therapist and orthodontist in Bangladesh. They are currently looking at establishing more specialist cleft center in other cities such as Khulna, Sylhet and Chittagong.⁵⁰

Erin, the Senior Vice President of programs visited the Rohingya refugee camp in Bangladesh to see the impact of Smile Train programs. With the influx of refugees into the area, she was visiting to speak with families, consult with our partners on the ground, and witness the impact of our partners firsthand and conducted a camp with the Smile train partners. They also found the heartbreaking stories of these patients. Cleft malnutrition is a global problem and need to keep under consideration while dealing with this issues.⁵¹

These factors are critical because patterns of medical and health-related service use and costs for patients with OFC can differ considerably by these factors. Additional studies are need to carry out to explore the costs of outpatient care, dental care, speech therapy, and special education, with consistent estimations of out-of-pocket expenses, and caregiver costs to determine the true economic burden of OFC in our country.

Understanding the effects of OFC and other craniofacial conditions on long-term health outcomes is important for quantifying the health burden and improving service delivery and healthcare policies for affected populations. However, much remains unknown about the effects of OFC on individual and family long-term health outcomes and on healthcare needs. One inherent limitation in conducting such studies has been the limited access to appropriate data sources and health registries that provide data individuals with OFC throughout life and provide large population-level random samples of unaffected individuals.

Currently, very limited information exists on the impact of OFC on indirect costs, such as loss in work productivity, time costs to parents, and effects of siblings' schooling. Understanding patterns of health service use and expenditures can help to target populations in need of services, assess the cost-effectiveness of treatments, and develop policies to improve the cost-effectiveness of and access to healthcare for patients and families with OFC.

II. CONCLUSION:

In recent times there has been a surge in research assessing the quality of life of individuals suffering from the different anomalies. Understanding the impact of oro facial clefts on these outcomes is important for identifying unmet needs and developing public policies to reduce the burden of oro facial clefts at the individual, family and societal levels.

Due to these considerations, the evaluation of patient's satisfaction from treatment, including possible associations between satisfaction from the esthetic and functional outcome with everyday life parameters, can offer valuable information to care providers. The investigation of the impact of a cleft on the social, professional, and family life of a patient relative to the esthetic and functional components of this specific condition may contribute to more successful and more targeted treatment approaches in the future.

ABBREVIATIONS:

CLP: Cleft lip and palate

OFC: Oro facial cleft

HRQOL: Health related quality of life

OHRQOL: Oral health related quality of life

LMICS: Low and middle income countries.

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