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Maternal Risk Factors and Family Burden with Cleft Children in Bangladesh

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ABSTRACT

Background: Congenital anomalies like cleft lip and palate are defects that are visible at birth and can be fatal. The family burden of cleft children is high in the world. **Methods:** This cross-sectional study included 110 cleft children (case) aged three months to 15 years. Data was collected through face-to-face interviews from parents or caregivers attended the Department of Oral and Maxillofacial Surgery of Dhaka Dental College and Update Dental College in Dhaka, Bangladesh. **Results:** This study included 58 (52.7%) males and 52 (47.3%) females cleft children. About 62 (56.4%) cases were <1 year old and the birth weight of 70 (63.6%) child was <2.5 kg. Fifteen (13.6%) mothers of them had a previous history of birth abnormalities with cleft children. Twenty-four (21.8%) mothers were malnourished during pregnancy. The mean caregiver burden score was 42.40; most were mothers, whereas 7 (43.8%) had no burden, followed by mild to moderate burden 13 (44.8%). Notably, 24 (36.9%) caregivers experienced moderate to severe burden levels with an age range of 30 to 40 years. Education up to HSC level and middle socio-economic status were found statistically significant for the birth of a cleft child ($p < 0.05$). **Conclusion:** These findings highlighted the significant impact on family burden and caregiver's well-being.

Practical Implication: This study aimed to determine maternal risk factors and the burden levels experienced by caregivers during the pre-operative phase at certain hospitals in Dhaka. This study will help to assess the cleft lip and palate among children.

Keywords: Bangladesh, Cleft lip, Cleft palate, Family burden, Cleft child

| **Submitted:** 10.08.2024 | **Accepted:** 18.09.2024 | **Published:** 21.09.2024

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How to Cite the Article

Latifa Howlader, Tanzima Yeasmin, Gias Uddin: Maternal risk factors and family burden with cleft children in Bangladesh.

IARJ. Med & Surg Res. 2024;5(5): 9-17.

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INTRODUCTION

Congenital malformations like cleft lip (CL), cleft palate (CP), and a combination of both cleft lip and palate (CLP) disrupt the core structures of the middle face and palate, significantly impacting patients' lives from birth. These deformities affect appearance, function, psychological well-being, and family and social interactions [1]. Globally,

orofacial cleft, including CL, CP, and CLP, affects approximately one in every 700 live newborns and varies in prevalence by race, with Asians (1 out of 440 live births). In Brazil, the reported prevalence is approximately one in every 650 live births. This condition presents a significant public health concern, affecting the quality of life for affected individuals and their caregivers [1-3]. The period of the child's growth and development relates to the palatoplasty procedure,

the main requirement of which is to modify the feeding habits of the child, including the food's consistency and the utensils used to feed them. The child at this age has developed eating preferences and techniques for consuming food [4]. Primary plastic surgery is crucial for repairing labio-palatal fissures in children [1].

Bottle use followed by cheiloplasty is still generally perceived. Its use following a palatoplasty- the most intrusive operative procedure among primary surgical operations- raises some controversy. Given their affordability, portability, and ease of cleaning, a glass and a spoon are reasonably practical substitutes [4]. During this time, caregivers should also be informed that the child should restart eating after coming out of the operating room, starting with a cold, liquid diet and continuing for 30 days [5]. Therefore, the condition and behavior of the child, as well as the care requirements associated with the therapy of labio-palatal fissures, may significantly impact the psychological and physical health of caregivers. Following surgery, caregivers face a higher risk of burden, exhaustion, social isolation, and increased stress due to the increased duty associated with the child's rehabilitation [6].

A surgery-related injury typically has a profound effect on the caregivers due to the varying degrees of stress brought on by the condition linked to the surgical risk, which can further exacerbate depersonalization disorder [7]. Objective caregiver load is defined as demands such as excessive duties, behavioral monitoring, financial difficulties, and disruptions. It is based on the frequency of help and supervision provided as part of a patient's daily routine care and the frequency of changes in the caregiver's social life. Various research has been conducted on the impacts of caregiver overload in various populations [8]. It is essential to recognize the burden by considering objective and subjective factors that can illustrate caregivers' physical and mental health, personal and social life, emotional well-being, and overall quality of life. Therefore, this study aimed to assess the level of burden felt by parents of children with cleft lip and palate.

MATERIALS & METHODS

Study Participants

This cross-sectional observational study was part of a large study including 110 cleft children. Cleft children were defined as cases aged three months to 15 years undergoing reconstructive surgery, along with their caregivers aged 18 to 50 years old. Caregivers of children with special cognitive, genetic, clinical issues, anomalies, or comorbidities were excluded. Cases were selected purposively, and face-to-face interviews collected data after obtaining informed verbal and written consent from each participant's caregiver and authority. The sampling technique aimed for a test power of 80%, 5% statistical significance, and a standard deviation with

a difference of 20% from the mean value of the Burden Interview (BI) [9].

Assessment of caregiver burden

The Burden Interview Scale (BI) was created by Zarit, Reever, and Bach-Peterson in 1980 to measure the burden that parents or unpaid caregivers of patients with mental, physical, or sensory impairments bear. The 22 items on this questionnaire covered the following topics: general well-being, financial status, social and personal life, emotional conduct, and interpersonal connections. Each answer had four possible scores: 0-never, 1-rarely, 2-sometimes, 3-often, and 4-always. The elements on the scale together gauged the extent to which the family caregiver felt overburdened by the child's care. Each item had four alternative responses and scores: 0-not at all, 1-a little, 2-moderately, 3-a lot, and 4 very much. The sum of all the scores gave the final BI score, which might vary from 0 to 100. The following methods were maintained during the analysis of the scores: 0-20 points predicted little or little burden, 21-40 points moderate burden, 41-60 points moderate to severe burden, and 61-88 points severe burden.

According to Zarit *et al.* (1980), the caregiver burden increases with the final score [10]. The BI scale, which measures caregiver stress for people with mental impairments, was translated and evaluated for the Brazilian population by Sczufca (2002). The Cronbach Alpha Coefficient yielded an index of 0.87, and the correlations between the replies for each item and the total number of questions were used to examine the psychometric aspects of the scale [9]. This study collected social and demographic data on children with craniofacial abnormalities. The research took place at two hospitals from August 2019 to October 2022, and data was gathered during the hospitalization of children undergoing surgery. Statistical analysis used the Chi-square test to examine the association between caregiver burden and variables such as number of children, gender, type of clefts, caregiver's marital status, educational achievement, and socio-economic position. IBM SPSS (Version-25) was used for data analysis with a significance level of 5% ($p < 0.05$ and $p < 0.01$) applied for every test.

Ethical consideration

The study obtained ethical permission from the Ethical Committee of the Institute of Biological Sciences, part of the University of Rajshahi. Both verbal and written informed consent was obtained from the study subject's caregivers.

RESULTS

A total of 110 children with cleft lips and palates were included in this study. Patients were selected from different districts attending Dhaka Dental College and Update Dental College in Dhaka. Table 1 shows 70 (63.6%) children

born with <2.5 Kg birth weight followed by current weight, 66 (60%) were in the age range of 4-10 Kg, 37 (33.6%) were the first child of the family. Among all, 68 (61.2%) showed feeding problems, 29 (26.4%) showed speech problems, and

61 (55.5%) of cases had nasal regurgitation. It also revealed that 15 (13.6%) had birth abnormalities with a cleft child and 2 (3.33%) had a history of an abnormal child in the family.

Table 1: Clinical characteristics of the clefts group (n=110)

Variables	Group	n = 60	%
Birth Weight (KG)	<2.5 kg	70	63.6
	2.5- 04 kg	31	28.2
	>04 kg	9	8.2
Present weight (KG)	<2.5 kg	0	0.0
	2.5-04 kg	15	13.6
	04-10 kg	66	60.0
	10-20 kg	29	6.67
	>20	22	20.0
Birth Order of child	1 st	37	33.6
	2 nd	14	23.3
	3 rd	31	28.2
	4 th	20	18.1
Associated Problems with CLAP Patients	Feeding Problems		
	Yes	68	61.2
	No	42	38.2
	Nasal Regurgitation		
	Yes	61	55.5
	No	49	44.5
	Speech disorder		
	Yes	29	26.4
	No	81	73.6
None	13	21.67	
Birth defects in cleft child	Absent	95	86.36
	Present	15	13.6
Abnormal child present in a family of cleft child	Yes	02	3.33
	No	58	96.67

n=frequency, %=percentages

Table 2: Position and types of clefts in study patients (n=110)

Characteristics	Frequency (n)	Percentage (%)
Site of Cleft lip and palate		
Unilateral Left sided	45	40.9
Unilateral Right sided	17	15.5
Median	29	26.4
Bilateral	19	17.2
Nature of Clefts		
Incomplete	42	38.2

Complete	68	61.8
Cleft Variations		
Unilateral cleft lip only (Incomplete)	19	17.3
Unilateral cleft lip and alveolus (Incomplete)	11	10
Unilateral cleft lip and palate (Complete)	32	29.09
Bilateral cleft lip only (Incomplete)	4	3.6
Bilateral cleft lip with alveolus (Incomplete)	2	1.8
Bilateral cleft lip and palate (Complete)	11	10
Median cleft lip only (Incomplete)	2	1.8
Median clefts with alveolus (Incomplete)	4	3.6
Median cleft lip and palate (Complete)	22	20
Facial clefts	3	2.7

Table 2 shows the position and types of cleft lip and palate

In Table 3 majority, 62 (56.4%) of cleft child were <1 year old. Among the cases, 58 (52.7%) were male and 52 (47.3%) were female. Besides, family history of cleft lip and palate was present in 8 cases (7.3%) which was significantly associated to cleft child ($p < 0.05$). About 82

(74.5%) mothers' age was between 20 to 35 years. Early marriage was about in 67 (61%) mothers with 31 (28.2%) were underweight. The majority, 46 (41.8%), lived in soil made house.

Table 3: Association between socio-economic demographic factors and cleft child (n=110)

Variables	Case (n=110)	Percentage (%)	P- value
Age group			
0-6 months	31	28.2	0.872
6 months- 1year	31	28.2	
01-05 year	27	24.5	
05-15 year	21	19.1	
Gender			
Male	58	52.7	0.787
Female	52	47.3	
Consanguineous Marriage			
Yes	42	38.2	0.823
No	68	61.8	
Maternal age(years)			
<20	27	24.5	0.874
20-35	82	74.5	
>35	1	1	
Paternal age(years)			
<20	8	7.2	0.892
20-35	87	79	
>35	15	13.8	
Early marriage			
Yes	67	60.9	0.784
No	43	39.1	
Maternal BMI			

Overweight and obese	13	11.8	0.737
Normal weight	66	60	
Under weight	31	28.2	
Family history of CLP			
Yes	8	7.3	0.035*
No	104	94.5	
Economic status			
Poor	51	46.4	0.893
Middle Class	59	53.6	
Residence			
Urban	41	37.3	0.679
Rural	69	62.7	
Type of house			
Tin Shed	28	25.5	0.738
Soil made	46	41.8	
Building	36	32.7	

P-value was determined by *Chi-square test
 P<0.01 was considered as strongly significant and p<0.05 was considered as statistically significant

vomiting in 1st trimester, and a history of taking folic acid in the first trimester were found to be significantly lower 61 (55.5%) and p value <0.001. Maternal nutritional deficiency was found to be significantly associated with birth of cleft child (p<0.001)

Table 4 stated that most of the mothers 76 (69.1%) didn't take proper diet and 64 (58.2%) had nausea and

Table 4: Association between maternal risk factors with the cleft children (n=110)

Attributes	n	%	P- value
Age group (in years)			
0-6 months	31	28.2	0.872
6 months- 1year	31	28.2	
01-05 year	27	24.5	
05-15 year	21	19.1	
Gender			
Male	58	52.7	0.787
Female	52	47.3	
Consanguineous Marriage			
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Tin Shed	28	25.5	0.738
Soil made	46	41.8	
Building	36	32.7	

P value was determined by *Chi-square test
 P<0.01 was considered as strongly significant and p<0.05 was considered as statistically significant

Table 5 stated that most of the mothers 76 (69.1%) didn't take

proper diet and 64 (58.2%) had nausea and vomiting in 1st trimester and a history of taking folic acid in the first trimester was found to be significantly lower 61 (55.5%) and p value <0.001. Maternal nutritional deficiency was significantly associated with cleft child birth (p=<0.001).

Table 5: Association between maternal risk factors with cleft children (n=110)

Maternal nutritional status	Case (n=110)		p-value
	n	(%)	
Proper diet in 1st trimester			
Yes	34	30.9	<0.001**
No	76	69.1	
Nutritional deficiency			
Yes	86	78.2	<0.001**
No	24	21.8	
Nausea and vomiting in 1st trimester			
Yes	64	58.2	0.498
No	46	41.8	
Folic acid taken in 1st trimester			
Yes	49	44.5	<0.001**
No	61	55.5	

P-value was determined by *Chi-square test.
 P<0.01 was considered as strongly significant and p<0.05 was considered as statistically significant

In Table 6, regarding the assessment of caregiver burden using the BI scale, a minimum score of 2 points and a maximum score of 61 were found. A total of 94 of the

caregivers showed evidence of burden. Most of them were mothers and presented a mild to moderate burden among 13 (44.8%) caregivers aged 30-40 year; moderate to severe in 32 (49.2%) caregivers of 29 years old. Education up to HSC level and middle socio-economic status were found statistically significant for birth of a cleft child.

Table 6: Association of caregiver's socio-demographic factors with burden status (n=110)

Attributes	Absent		Mild to moderate		Moderate to severe/severe		p-value*
	n	%	n	%	n	%	
Primary Caregiver							

Mother	14	87.5	26	89.7	61	93.8	0.431
Father	2	12.5	2	6.9	2	3.1	
Grand parents	0	0	1	3.4	2	3.2	
Caregiver's age (in years)							
18 to 29	8	50	12	41.4	32	49.2	0.898
30 to 40	7	43.8	13	44.8	24	36.9	
>40	1	6.3	4	13.8	9	13.8	
Education							
Below HSC	6	37.5	16	55.2	17	26.2	0.031*
HSC	9	56.3	12	41.4	33	52.3	
Graduation	1	6.3	3	3.4	14	21.5	
Socio-economic Status							
Low	0	0	12	55.2	31	47.7	<0.001*
Middle	9	56.3	16	41.4	34	52.3	
High	7	43.8	1	3.4	0	0	

P-value was determined by Fisher Exact test. $P < 0.01$ was considered as strongly significant and $p < 0.05$ was considered as statistically significant

DISCUSSION

The current study was carried out to evaluate the epidemiological characteristics of orofacial clefts. A total of 110 children with cleft lip and palate were enrolled in the cross-sectional survey at Dhaka Dental College & Hospital, Update Dental College Hospital in Dhaka. The prevalence of cleft children varies across ethnic groups and is thought to be higher in developing nations. Identifying the epidemiological traits of these patients and their unique characteristics is essential to provide a more effective multidisciplinary treatment algorithm. The prevalence of cleft lip and cleft palate has increased in recent years. However, these epidemiological figures vary according to the geographical region and each population's demographic and social characteristics, even within the same country and locality.

In the current study, most participants in the patient's group were under one-year-old 62 (56.4%). A previous study by Solano *et al.* demonstrated that the largest age distribution of patients with clefts was found in neonates, followed by infants younger and older up to 1 year [11-13]. The male-female ratio is consistent with the previous study, which stated that cleft was more frequently found in male patients (58 (55.95%)) than females (44.05%) [14]. Kishimba *et al.* (2015) also observed a slight male predominance among the cases [15]. The gender distribution of the sample was uniform, with males being more likely to have labio-palatal fissures and girls more likely to have an isolated cleft palate (CP) [16]. The infants underwent cheiloplasty and palatoplasty procedures, indicating that the procedure involving primary surgeries according to the child's age was followed. Trettene *et al.* also found a similar outcome [4]. The children were 1 year old on average, and labio-palatal fissures were more common. If the operation were performed at the right moment, it would help

in a variety of ways to ensure that the child with a labio-palatal fissure has a successful rehabilitation process [17].

According to the current study, the birth weights of the cases were low; about 70 (63.6%) weighed 2.5 kg. Besides this, a family history of the abnormal child was present in 2 (3.3%) of cases. A previous study also revealed that low birth weight babies were almost four times more likely to have structural congenital disabilities compared to normal-weight babies [15]. Some other studies also observed that family history had a positive association with clefts. 13,16 Approximately 76 (69.1%) of mothers didn't take a proper diet, and 64 (58.2%) had nausea and vomiting in the first trimester. About 61 (55.5%) did not take folic acid in the first trimester properly. There was a higher prevalence of 86 (78.2%) cases of maternal nutritional deficiency.

Nutritional fortification and supplementation significantly impact controlling congenital disabilities as a public health measure. Low socio-economic status predisposes women of reproductive age to an increased risk of malnutrition before and during pregnancy [15]. The majority, 62 (56.4%) had unilateral, 29 (26.4%) had a median type, and 19 (17.2%) had bilateral cleft lip and palate. Sixty-eight (61.8%) of cases were complete type, while 42 (38.2%) of the cleft lip cases were incomplete. Among them, unilateral incomplete was 30 (27.3%) cases, and facial clefts were 3 (2.7%) cases. In the study of Solano *et al.*, 79.5% of cases were unilateral CL, and 48.9% were incomplete [13]. Another study also observed that most had unilateral cleft lip and palate followed by bilateral and median cleft lip and palate [18].

About 42 (38.2%) caregivers had a history of consanguineous marriage. Though no significant association was found, long-term study with large sample size may reveal the exact scenario. Regarding caregiving of the cases, more than 90% were mothers (101), followed by fathers and

grandparents. The mean caregiver burden score was 42.40; most were mothers, whereas 7 (43.8%) had no history of burden, followed by mild to moderate burden 13 (44.8%). Notably, 24 (36.9%) caregivers experienced moderate to severe burden levels with an age range of 30 to 40. Socio-demographic characteristics of the caregivers and low educational and economic status were significantly associated with burden status ($p < 0.05$). High socio-economic status showed significantly lower burden scores. The previous study showed that the majority of the case's primary caregiver was the mother and also demonstrated the levels of caregiver burden as severe (4.4%), moderate to severe (21.1%), mild to moderate (40%), and little or none (34.5%). In contrast, the low economic condition was the only significant and independent predictor of caregiver burden. Moreover, the caregivers' age, level of education, and employment status were not significantly associated with caregiver burden [19]. In a previous study, 23.2% of caregivers declared a high level of burden, 49.1% declared an average level of burden, and 27.7% declared a low level. In contrast, higher education levels have been reported to be associated with lower caregiver burden in previous studies [20].

The impact factors that influence the level of burden can be measured, and effective programs to support parents can reduce the burden level. The study was cross-sectional observational, and the sample was collected by purposive sampling. These were important limitations of this study.

CONCLUSIONS

In our sample, more males than females had cleft lips and palates. Patients with cleft lip and palate in developing countries might not seek medical assistance because they are unaware of the treatment. Bangladesh must implement a birth surveillance system for congenital abnormalities to support comprehensive treatment for individuals with CLP and inform health service planning and policy. Consequently, to provide children with adequate therapy for special needs, mainly when those needs are being treated in a hospital, public and private institutions that provide primary healthcare must consider the mental and physical health of the caregivers.

Acknowledgments

The authors like to express their gratitude to the employees and the outpatient clinics with respondents at Dhaka Dental College & Hospital and Update Dental College & Hospital.

Author Contributions

Latifa Howlader designed the study, collected and analyzed data, and drafted the manuscript. Tanzima Yeasmin supervised the research and contributed to data interpretation and manuscript review. Gias Uddin assisted with data acquisition and manuscript revision. All authors approved the final manuscript and ensured its accuracy and integrity.

Ethics Statement

The Ethical Committee of the Institute of Biological Sciences, University of Rajshahi (Memo No. 73 (01)/320/IAMEBBC/IBSc) approved the study. Informed verbal and written consent was obtained from all participants' caregivers before data collection. The study was conducted under the Declaration of Helsinki.

Conflict of Interest Disclosures

The authors declare no financial relationships or conflicts of interest related to this study. No financial support was received for the submitted work.

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