Factors Responsible for Unfavorable Dental Arch Relationship in non Syndromic Unilateral Cleft Lip and Palate Children

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Objectives: *Multiple factors are whispered to be crucial cause of unfavourable dental arch relationship in cleft lip and palate (CLP).This study aims to evaluate the dental arch relationship of Bangladeshi children with non syndromic unilateral cleft lip and palate (UCLP) following cheiloplasty and palatoplasty. Also to explore the various congenital (UCLP type, UCLP side, family history of cleft, family history of class III) and environmental (cheiloplasty, palatoplasty) factors that affects dental arch relationship of UCLP patients.* ***Study design:*** *This was a retrospective study where 84 dental models were taken before orthodontic treatment and alveolar bone grafting. The mean age was 7.69± 2.46 (mean± SD). The dental arch relationship was assessed by GOSLON (Great Ormond Street, London and Oslo) Yardstick. According to GOSLON Yardstick, five categories are rated; named- 1: excellent; 2: good; 3: fair; 4: poor; 5: very poor. Also the groups have been dichotomized into favorable (category ratings 1-3) and unfavorable (category ratings 4 and 5) groups. Kappa statistics was used to evaluate the intra- and inter-examiner agreements and logistic regression analysis was used to explore the responsible factors that affect dental arch relationship.* ***Results:*** *Total 37 subjects (44% of all subjects) were categorized into unfavourable group (category rating 4 and 5) using GOSLON yardstick. Intra- and inter-examiner agreements were very good. The mean GOSLON score was 3.238. Using crude and stepwise backward regression analysis, significant association was found between family history of skeletal class III malocclusion (p = 0.015 and p = 0.014 respectively) and unfavourable dental arch relationship. Complete UCLP (p = 0.054) and left sided UCLP (p = 0.053) also seemed to be correlated but not significant with unfavourable dental arch relationship using crude and stepwise backward regression analysis respectively.* ***Conclusion:*** *This analysis suggested that family history of skeletal class III was significantly correlated with unfavourable dental arch relationship of Bangladeshi UCLP children.*

Key words: *Unilateral cleft lip and palate; Dental arch relationship; GOSLON Yardstick.*

List of Abbreviations:*Cleft lip and palate (CLP); Unilateral Cleft Lip and Palate (UCLP).*

INTRODUCTION

Any deformities (anatomical or chromosomal) that instigated during pregnancy and their effects have been detected after birth considered as congenital anomalies.[1] Among all, cleft lip and palate (CLP) is one of the most common congenital malformations[2] that can occur together or individually. The etiology of CLP is still controversial. According to previous studies, it is to be thought that both major and minor genetic with variable environmental factors are responsible for CLP.[3] Multifarious functional problems like feeding, speech, hearing, dental functioning and also psychological dilemma can happen to the patient. Mid face deficiency, maxillary arch constrictions, congenitally missing and malformed teeth, orthodontic anomalies like crowding, rotation, malposition of teeth are frequently observed in CLP patient.[4] CLP shows different prevalence in different civilization and races in addition to countries. In Asian population, CLP affects approximately 1.30 of every 1000 live births.[5] Moreover the prevalence rate of USA is 2.2 to 11.7 per 10,000 live births[6].

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Several indices such as the GOSLON(Great Ormond Street, London and Oslo) Yardstick[7], the 5-year-old index[8], the GOAL (Goteborg (G), Sweden; Oslo (O), Norway; Aarhus (A), Denmark; and Linkoping (L), Sweden) index[9], EUROCRAN index[10], Huddart/ Bodenham scoring system[11], modified Huddart Bodenham scoring system[12, 13] are used to assess dental arch relationship in patients with CLP. Specific index has its individual uses and advantages. However, The GOSLON Yardstick is the most frequently used index which rated the malocclusions according to antero-posterior arch, vertical labial segment, and transverse relationships in patients with UCLP.[14]

Spectrum of factors influence treatment outcome such as UCLP type, affected side, family history of cleft and class III malocclusion, chieloplasty, palatoplasty and auxiliary intervention etc. Lack of consideration of factors affecting outcome of treatment in children with CLP has led to great diversity of protocols and surgical techniques working by various cleft groups worldwide.[15-20]As a result to perceive level of treatment outcomes, the progress of methods is required if surgeons are to surround a sound basis on which they can justify modifications of their timing or techniques.[21]

In contemporary era, multitude of research on CLP has been done worldwide.In a typical developing country like Bangladesh, more than 5000 CLP patients are born every year in Bangladesh where the prevalence rate is 3.9 *per* 1000 live births.[22] But according to literature survey no clinical study up till now has been done in Bangladesh regarding dental arch relationship and treatment outcome of UCLP patients. Various researcher evaluated treatment outcome based on individual factors.[17-19] But, very few researches has been done considering various factors at a time to explore the responsible factor that affect dental arch relationship in UCLP children.[23, 24]The aim of the study was to evaluate the dental arch relationship of Bangladeshi children with non syndromic unilateral cleft lip and palate (UCLP) following cheiloplasty and palatoplasty and to explore the responsible congenital and environmental factors that affect dental arch relationship of UCLP patients using GOSLON Yardstick.

MATERIALS AND METHOD

This study was approved by the Ethical Committee of the Hospital Universiti Sains Malaysia (HUSM) [USM/JEPem/15020039], which complies with the Declaration of Helsinki. The dental arch relationship was examined among 84 dental models of non syndromic UCLP children.Among the selected subjects, 43 subjects were male and 41subjects were female. Fifty one patients had left sided UCLP. Thirty one patients had complete UCLP.There were 50 subjects had a family history of cleft and 34 subjects had family history of skeletal class III malocclusion (mandibular prognathism and/or maxillary retrognathism) (Table 1). All subjects had undergone cheiloplasty at the average age of 5 months. In 35 subjects, Milliard technique for lip closure had been performed and in 49 subjects, modified Milliard technique had been performed (Table 1). All subjects underwent palatoplasty at the average age of 18 months. Forty four subjects underwent Bardach technique of palatoplasty and 40 subjects underwent V-Y pushback technique (Table 1). All the models, history and examination record of patients were achieved from an archive of a renowned hospital in Bangladesh.

Table 1.Distribution of subjects with variable factors.

Variables	Number
Gender	
Male	43
Female	41
UCLP affected side	
Right	33
Left	51
UCLP types	
Complete	31
Incomplete	53
Family history of cleft	
Positive	50
Negative	34
Family history of Class III	
Positive	34
Negative	50
Palatoplasty	
Bardach technique	44
V-Y pushback technique	40
Cheiloplasty	
Modified Millard technique	49
Millard technique	35

Sample size calculation

To study prevalence of successful treatment outcome using GOSLON Yardstick,

$$n = \{Z/\Delta)^2 \times P (1-P)$$

Where Z = 1.96 (level of significance = 0.05)

Absolute precision, Δ = 0.10 (10%) and Anticipated population proportion, P = 0.317

Width of	
Δ	N
0.40/0.16	
0.35/0.1225	
0.30/0.09	9
0.20/0.04	21
*0.10/0.01	84

If the absolute precision 10%, the sample size required is 84.

For logistic regression the sample size is estimated by using a ratio 1 predictor: 12 cases. In our study there are seven predictors. Thus 84 cases are required.

Inclusion criteria were:

Non syndromic UCLP patient. Individuals aged 5-12 years. Cheiloplasty and palatoplasty had been performed. No alveolar bone graft. No orthodontic treatment.

Exclusion criteria were:

Subjects with bilateral CLP and isolated cleft palate. Syndromic UCLP. Cheiloplasty and palatoplasty had not been performed. Bone grafting had been done. Orthodontic treatment had been started.

GOSLON Yardstick[7, 23, 25] was used to evaluate dental arch relationship. According to GOSLON Yardstick, five categories are rated; named- 1: excellent; 2: good; 3: fair; 4: poor; 5: very poor which reflect a growth range of dental arch relationship. Group 1

(excellent), a favorable relationship, shows advantageous skeletal form, with a positive overjet and overbite. Patients exhibit an Angle class II division 1 malocclusion in this group. Straightforward orthodontic treatment or no treatment need at all in this group. Group 2 (good) is also a favorable relationship with Class I dental relationship and also indicates straightforward orthodontic treatment. Group 3 (fair) presents as an edge-to-edge dental relationship where patient need of more complex orthodontic treatment to correct the Class III malocclusion and other possible arch deformities, but a good result can still be predictable. Group 4 (poor), an unfavorable facial growth with reverse overjet of 3-5 mm which indicates the limits of orthodontic treatment, may require an orthognathic procedure. Group 5 (very poor) represents a significant skeletal class III relationship with mandatory surgical correction.

Numbers were randomly assigned to each model by simple random sampling technique, and no other form of identification was visible. Five examiners rated the 84 models of UCLP subjects twice with two weeks interval. Taking together the data in each model, we generated a mean score.[26] The subjects were divided into two groups; favorable (category ratings 1-3) and unfavorable (category ratings 4 and 5) groups. This grouping was carried out because the patients in the favorable groups could be treated with conventional orthodontics, whereas patients in the unfavorable groups sometimes required surgical correction.[25]

Statistical analysis

The intra- and inter-examiner agreements wereanalyzed with the kappa statistics. According to Altman[27], the kappa values of the intra- and inter-examiner agreements were interpreted. Various factors with favorable and unfavorable outcomes were evaluated by Chi square test. Logistic regression analysis was performed using the dichotomous dependent variable, favorable and unfavorable groups. Both crude and backward stepwise logistic regression analyses were done to explore the unfavorable dental arch relationship in UCLPpatients. These analyses were carried out using the statistical package SPSS Version 22.0 (SPSS Inc., Chicago, IL, USA). Significance level was set at $p < 0.05$.

RESULTS

Intra- and inter-examiner agreements

Intra-examiner agreements for examiner A, B, C, D and E were 0.873, 0.888, 0.904, 0.870 and 0.856 (Table 2). The kappa score ranged from 0.809 to 0.951 for the inter-examiner(Table 2). The kappa scores for the GOSLON Yardstick showed very good intra- and inter-examiner agreements.

Among the 84 subjects, scores were distributed as follows: category 1= 2 subjects, 2=19 subjects, 3=26 subjects, 4=31 subjects and 5= 6 subjects. The mean GOSLON score was 3.238 (Figure 1).

Table 2.Intra- and inter-examiner agreements.

Intra-examiner	Kappa value	Standard error
A	0.873	0.043
B	0.888	0.041
C	0.904	0.038
D	0.870	0.044
E	0.856	0.045
Inter-examiner		
First rating		
A vs. B	0.889	0.040
B vs. C	0.920	0.035
C vs. D	0.951	0.028
D vs. E	0.935	0.032
E vs. A	0.920	0.035
Second rating		
A vs. B	0.809	0.051
B vs. C	0.840	0.048
C vs. D	0.936	0.031
D vs. E	0.904	0.038
E vs. A	0.809	0.051

Comparison of factors between favorable and unfavorable groups

Distribution of the percentage of favorable and unfavorable group of various factors like sex (P value 0.179), UCLP type (P value 0.128) , UCLP side (P value 0.809), family history of cleft (P value 0.662), family history of class III malocclusion (P value 0.024), type of cheiloplasty (P value 0.795), type of palatoplasty (P value 0.543) are given in figure 2.

Crude logistic regression analysis

Crude logistic regression analysis was carried out to quantify the strength of association between each factors and dental arch relationship. The 95% confidence intervals were determined and the factors with a p-value of less than 0.05 were considered to have a significant association with dental arch relationship. Significant association was found between family history of Class III (*p*value= 0.015) and unfavorable dental arch relationship. Cheiloplasty with Milliard technique (odds ratio= 1.442) is also leading to unfavorable dental arch relationship and incomplete UCLP is leading to favorable dental arch relationship since their odds ratio is higher (>1) and lower (>1) respectively (Table 3).

Stepwise logistic regression analysis

Stepwise logistic regression analysis planned to explore the association between various factors (independent variable) and dental arch relationships (dependent variable). The 95% confidence intervals were determined and the factors with a p-value of less than 0.05 were considered to have a significant association with dental arch relationship. Family history of Class III (*p* value= 0.014) showed significant association with unfavorable dental arch relationship. Right sided UCLP (*p* value= 0.053) also seemed to be correlated with favorable dental arch relationship but no significant association was found (Table 3).

Figure 1. Score distribution (percentages) for 84 UCLP subjects using GOSLON Yardstick. The mean GOSLON score was 3.238.

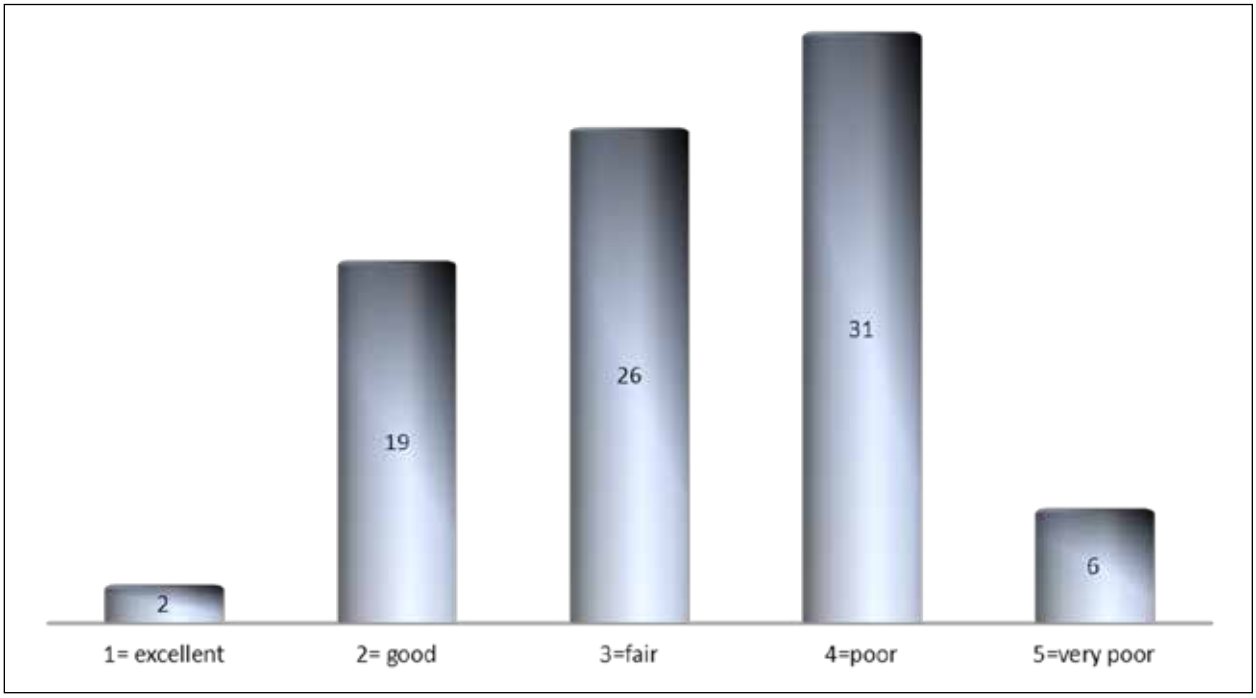

Figure 2. Distribution of subjects with variable factors in favorable and unfavorable groups using GOSLON Yardstick (the number of subjects in favorable and unfavorable groups was 47 and 37, respectively).

CUCLP: Complete UCLP,ICUCLP: Incomplete UCLP, FH cleft +ve: Positive family history of cleft, FH cleft –ve: Negetive family history of cleft, FH Class III +ve: Positive family history of Class III malocclusion, FH Class III –ve: Negative family history of Class III malocclusion, Cheiloplasty-MMT: Modified Millard technique of chieloplasty, Cheiloplasty-MT: Millard technique of chieloplasty, Palatoplasty BT: Bardach technique of palatoplasty, Palatoplasty V-Y PT: V-Y pushback technique of palatoplasty.

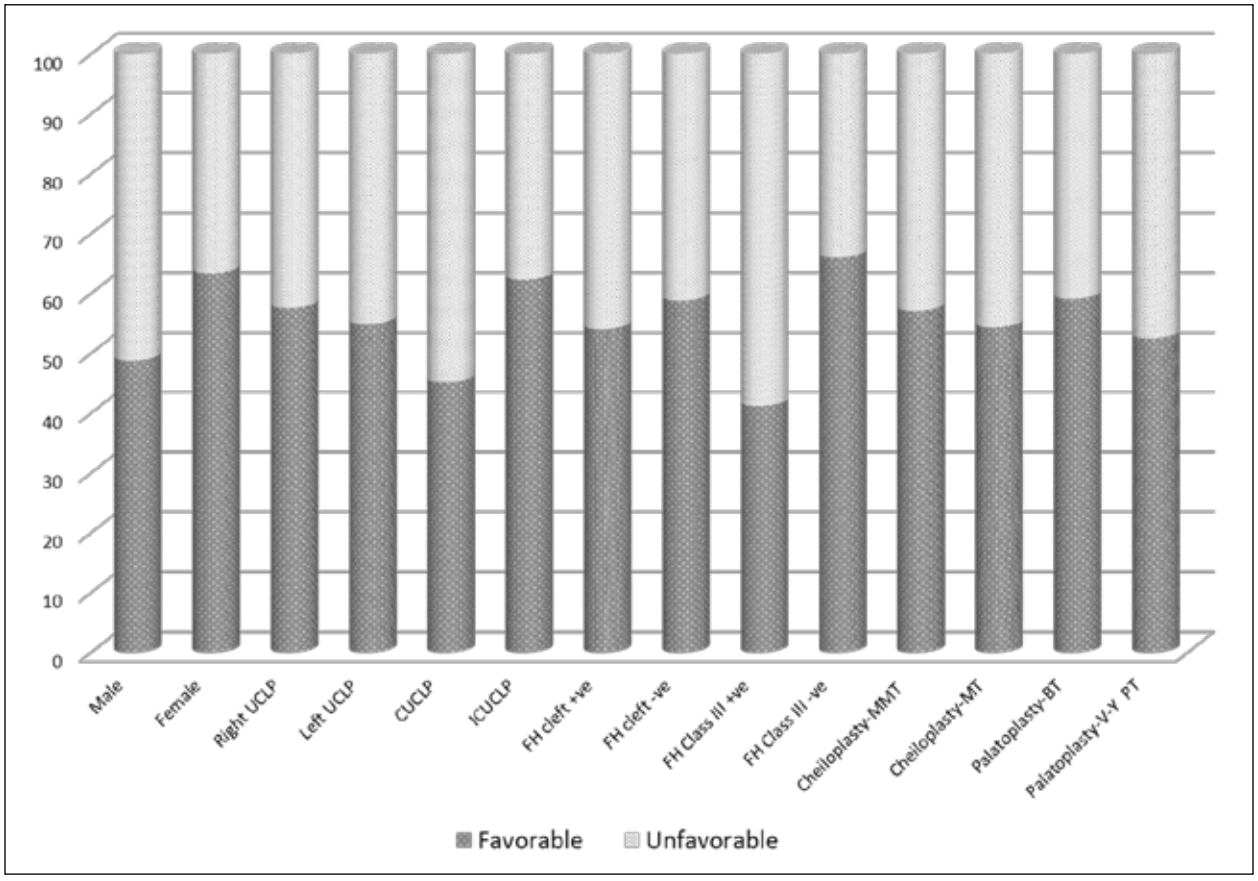

Table 3. Crude odds ratio and adjusted odds ratio (stepwise regression analysis: backward method): favorable vs. unfavorable group using GOSLON Yardstick.

Crude logistic regression analysis			
Variable	**Odds ratio**	**95% confidence interval**	**P Value**
Age	1.098	0.897-1.348	0.364
Gender (male)	0.547	0.209-1.431	0.219
UCLP affected side (right)	1.117	0.401-3.110	0.832
UCLP type (incomplete)	0.199	0.039-1.026	0.054
Family history of cleft (+ve)	1.004	0.342-2.946	0.994
Family history of Class III (-ve)	0.285	0.103-.785	0.015
Palatoplasty with Bardach technique	0.465	0.068-3.189	0.436
Cheiloplasty with Milliard technique	1.442	0.369-5.638	0.595
Stepwise logistic regression analysis:			
UCLP affected side (right)	0.215	0.045-1.023	0.053
Family history of Class III (-ve)	0.303	0.117-.784	**0.014**
Palatoplasty with Bardach technique	0.386	0.085-1.748	0.217

An odds ratio greater than 1 indicates that the respective independent factor associates with unfavorable dental arch relationship, and less than 1 indicates that the respective independent factor associates with favorable dental arch relationship

DISCUSSION

Over the last two decades, Goslon Yardstick index[7] is observed as the most commonly used index.[28]In this prime study, we assessed 84 models of non syndromic UCLP subjects for evaluation of dental arch relationship using GOSLON Yardstick. GOSLON Yardstick revealed itself to have good inter- and intra-examiner reproducibility and reliability.[29] Categorizing dental arch relationships and interfering of facial morphology outcomes, GOSLON Yardstick has proved its capability between different centers.[30] It can also predict surgical outcomes at early age of patients[31] as well as it is also associated with results of cephalometric analysis.[23]The treatment outcome of the most subjects of our study was fair to poor, representing 68% of all cases. Of the left over, 23 % was good and 2 % was excellent prognosis and 7 % was very poor outcome. According to literature survey, no studies had been done regarding CLP about Bangladeshi population; it is not possible to compare the results of this study with previous study about this population. However, there have been many studies done about CLP with GOSLON Yardstick in other population. For example, in a study of Japanese population by Alam *et al.,*[24] found fair to poor outcome (GOSLON 3 and 4) in 80% of the UCLP patients. Similar study was performed in Finland population by Harila*et al,*[32] found good (GOSLON 1 and 2) in 77.1% of all cases. Dogan*et al,*[33] deliberated Turkish population and established 50.4% of the patients were scored as unfavorable GOSLON. Correspondingly Sinko *et al,*[34] found favorable GOSLON score in 71.5% of the UCLP cases. Different population showed different results. These may be due to uses of different technique of surgery and/or the experience of the surgeons.

This study focused on the possible effects of various congenital (UCLP type, UCLP side, family history of CLP, family history of class III malocclusion) and environmental (cheiloplasty, palatoplasty) factors on dental arch relationships. To observe the associations between each congenital and environmental factor and dental arch relationships,crude logistic regression analysis were carried out. And Stepwise logistic regression analysis was carried out to explore the associations between factors and dental arch relationships. By using both crude and backward stepwise logistic analysis, the results of this current study revealed that the subjects who had no family history of class III malocclusion showed favorable dental arch relationship; that means the positive family history of class III malocclusion significantly affect the dental arch relationship. In a study, similar findings have been reported by Alam *et al*.[24] They also found family history of class III malocclusion is correlated with dental arch relationship in a Japanese population.

This study demonstrated that subjects who had complete UCLP are more likely to have unfavorable dental arch relationship. Moreover, we found subjects who had left UCLP are more likely to have unfavorable dental arch relationship.

In this retrospective study, all cheiloplasty and palatoplasty were executed at the same hospital and operated by two different surgeons by utilizing same treatment protocol just applying two different techniques of surgery. As a result, we could evaluate the techniques of surgery which was responsible for unfavorable dental arch relationship. It is interesting to note that patients who were lip repaired with Millard technique had favorable dental arch relationship than modified Millard technique but not significant, although this variable did not reach as a precise factor stepwise regression analysis. In some other studies, researchers suggested that modified Millard technique is more favorable than modified Millard technique with vomar flap.[23, 24]. On the other hand, Apostol[35] revealed Onizuka technique was not only satisfactory to the patients but also to the surgeon concerning the esthetical and functional purpose. Meyer and Seyfer[36] found Tennison technique presented more flexibility with wide clefts and Millard technique presented outstanding results with narrow clefts.

However, concerning palatoplasty, till few years back, V-Y pushback technique of palatoplasty was one of the familiar techniques though after surgery a widespread raw surface is produced both anteriorly and laterally causes shorten of palate[37] which leads to unfavorable dental arch relationship. In this study we found subjects who underwent V-Y pushback technique of palatoplasty showed poor prognosis than the subjects who underwent palatoplasty with Bardach technique of palatoplasty. Johnston *et al,*[38] found most of the patients who underwent V-Y pushback palatoplasty scored GOSLON 4 and 5 (needed orthognathic surgery) though statistically they did not found any significant differences. Abdel-Aziz and Ghandour[39] executed a comparative study between V-Y pushback technique and Furlow technique and reported that treatment outcome (velopharyngeal adequacy and speech outcome, fewer probability of palatal fistula) of Furlow technique showed better prognosis than V-Y pushback technique. But in another study Jain *et al,*[40] found V-Y pushback technique had good prognosis for speech. Until today, there is no consensus on which surgical technique is the best in view of the outcome of

surgery either for lip repair or for palate repair. The differences of the severity among cases, the aim of the surgery, the surgeons experience, expertise and preferences may affect the outcome of the surgery as well.

From the results of this study, we found that family history of class III malocclusion, complete UCLP are the predictors of unfavorable dental arch relationship in young children with UCLP. Moreover, UCLP sides, type of cheiloplasty, type of palatoplasty are also somewhat responsible for the favorable and unfavorable dental arch relationship.

These findings were achieved from Bangladeshi UCLP subjects (Model) assessed by GOSLON Yardstick. May be these findings are different in other population. We encourage other population to do same study to explore the precise factors that are responsible for dental arch relationship. In future, longitudinal research is planned. Furthermore, we plan to explore the congenital and/or environmental factors affecting craniofacial morphology using cephalometric analysis of Bangladeshi UCLP subjects.

CONCLUSION

The present study shows that,

1. in Bangladeshi UCLP subjects, the mean score of GOSLON Yardstick was 3.238.
2. this study revealed that there was a significant association between family history of skeletal class III malocclusion and unfavourable dental arch relationship using crude and stepwise regression analysis.

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