

# Dental Arch Relationships of Saudi Children with Unilateral Cleft Lip and Palate

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## ABSTRACT

**Aim:** To determine dental arch relationships of Saudi children born with nonsyndromic complete unilateral cleft lip and palate (UCLP).

**Material and methods:** This is a retrospective cohort study that comprised dental study models of 74 UCLP Saudi children aged 8–10 years who were recruited from 14 referral cleft centers. All participants had their cleft lip and palate repaired with no history of alveolar bone graft or any orthodontic treatment. Dental arch relationships of UCLP patients were assessed using the Great Ormond Street, London, and Oslo (GOSLON) Yardstick—a clinical tool that categorizes dental relationships of UCLP children into five discrete grades from I to V. The reliability of the rating was assessed with weighted kappa ( $\kappa$ ) statistics.

**Results:** Three children (4.1%) had excellent surgical outcomes (grade I), 18 children (24.3%) filled into grade II (good outcome), 22 subjects (29.7%) had grade III (fair outcome), 27 children (36.5%) had grade IV (poor outcome), and 4 subjects (5.4%) were ranked as having very poor outcomes (grade V). The mean GOSLON score was 3.39. Intrarater and interrater agreements were high indicating good reproducibility.

**Conclusion:** Based on the dental arch relationships, the treatment outcome of UCLP Saudi children was unsatisfactory, with a mean GOSLON score of 3.39. Delayed palate repair and the use of presurgical orthopedics may be considered in the future for cleft deformity management.

**Clinical significance:** To address the effect of particular cleft surgical protocol on dental arch relationships of UCLP patients.

**Keywords:** Dental arch relationships, GOSLON Yardstick, Unilateral cleft lip and palate.

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## INTRODUCTION

Cleft lip and palate is a common congenital craniofacial deformity that affects one out of every 700 newborns worldwide.<sup>1</sup> This deformity can have a significant impact on facial esthetics, speech, feeding, and psychosocial wellbeing.<sup>2</sup> The primary objective of cleft lip and palate repair is to restore and optimize midfacial development, articulation, and psychological and social integration of children born with cleft lip and palate. Individuals with uncorrected CLP tend to develop normal growth patterns in their craniofacial complex.<sup>3,4</sup> The greatest growth inhibitory effect was observed following 1-stage palatal repair according to Veau–Wardill–Kilner protocol, when performed at 12 months of age while the least amount of facial retrusion and discrepancy in development of the dental occlusion was noted in the 2-stage operation of soft palate repair at 1–2 years and hard palate repair after age 12.<sup>5</sup> However, delay of cleft surgery may negatively impact speech and related quality of life parameters. The controversy of appropriate timing and technique for cleft surgery has been a topic of ongoing discussion over the years.

The dental arch relationship is a critical component of the overall facial esthetics and function in individuals with cleft lip and palate. Several indices have been developed to evaluate and quantify the severity of dental arch discrepancies in these patients. Pruzansky and Aduss,<sup>6</sup> and Matthews et al.,<sup>7</sup> proposed methods based on the presence and extent of crossbite. However, these were extremely outdated and did not accurately describe the extent and severity of malocclusion. Another index used was the Huddart–Bodenham (H–B) system,<sup>8</sup> which evaluates dentoalveolar relationships in deciduous dentition based on the location of the most posterior tooth in relation to the occlusal plane. The modified H–B system is another index that has been developed to overcome the limitations of the original H–B system.<sup>9</sup> It combines a visual assessment of

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the dental arch relationship with an objective measurement of the location of the most posterior teeth utilizing a 40-point scale. According to WHO, the modified Huddart–Bodenham scale had the best performance in ranking. However, the GOSLON Yardstick is the most commonly used index with a prevalence of more than three times the rest of the indices combined.<sup>10</sup>

Great Ormond Street, London, and Oslo (GOSLON Yardstick) is a standardized index that was developed to assess dental arch relationships in children with unilateral cleft lip and palate (UCLP) in late mixed and early permanent dentition.<sup>11</sup> The index ranks dental arch relationships based on five categories, ranging from grade I (excellent) to grade V (very poor), with grade V representing the most severe malocclusion. The GOSLON Yardstick allows for consistent and objective assessment of dental arch relationships in children with UCLP and has been shown to have high inter- and intraexaminer reliability, making it a reliable tool for clinical and research use.

Saudi Arabia has a prevalence of cleft lip and palate, estimated at 1.17 per 1000 live births making it marginally lower than the mean global prevalence. Unilateral cleft lip and palate is the most common type, accounting for approximately 70% of cases.<sup>12</sup> Interestingly, Zamzam water use was associated with a reduced risk of developing cleft lip and palate. Zamzam water contains a lot of minerals which is not available in bottled and tap water and it is only found in the Makkah region of the country.<sup>13</sup> In recent years, Saudi Arabia has developed a well-established network of cleft centers that provide multidisciplinary care for children with cleft lip and palate, including surgical, orthodontic, and speech therapy services. However, the database of cleft deformity in Saudi children is still lacking. The purpose of this paper is to examine the dental arch relationship in non-syndromic UCLP in Saudi Arabia using the GOSLON Yardstick.

## MATERIALS AND METHODS

This retrospective study was approved by the Research and Ethics Committee of the Dental School at the University. This study comprised study models of consecutive Saudi children with complete UCLP who were born between 1 January 2010 and 1 January 2014 (8–10 years). Study models were identified and recruited from 14 dental schools and orthodontic departments of hospitals at Ministry of Health. Reviewing the records of the participants revealed that all cleft surgery was performed by the 11 plastic surgeons following the same surgical protocol; lip closure was undertaken at 6 months using the modified Millard rotation advancement; flaps from either side of the cleft lip and nasal base are elevated. Then, surgeons rotated and advanced the flaps to close the cleft and create a natural lip contour. On the contrary, the palate closure was performed at 18 months using a “V–Y pushback” technique where two flaps of mucosa on either side of the cleft are elevated and released from the bone and bulb of the palate allowing the integrity of the palate. No presurgical orthopedics, alveolar bone graft, or any active orthodontic treatment was undertaken in any subject at the time of assessment. Study models of poor quality, those operated with different surgical protocols, and syndromic cleft cases were excluded from the study.

Orthodontic study models were collected and numbers were then randomly assigned to each model and marked in pencil with no other means of identification. Assessment was performed by two orthodontists (A and B) who were trained in using GOSLON Yardstick. Both assessors rated the Yardstick twice to examine the reliability and reproducibility of the ranking.

### Statistical Analysis

Linear weighted kappa ( $\kappa$ ) was used to assess intra- and interexaminer agreement. The test is a statistical measure of inter-rater agreement or reliability for categorical items where the values are ordered. It assesses the degree of agreement among raters, accounting for chance agreement. The  $\kappa$ -score ranges from 0 to 1, with a value of 0.60 indicating good agreement while more than 0.80 indicates a very good strength of agreement. In addition, an independent *t*-test [Statistical Package for the Social Sciences (SPSS), version 20.0, software, Chicago, Illinois, USA] was used to detect differences in cleft side and gender distribution among the sample.

## RESULTS

Table 1 shows the demographic features of Saudi UCLP children. The mean age of the UCLP children was 9.17 years (SD, 1.76 years).

**Table 1:** Demographic features of Saudi UCLP children

	Left UCLP (%)	Right UCLP (%)	Total (%)
Male	28 (37.8)	12 (16.2)	40 (54.0)
Female	25 (33.8)	9 (12.2)	34 (46.0)
Total	53 (71.6)	21 (28.4)	74 (100)

**Table 2:** The GOSLON Yardstick ranking of Saudi UCLP children

GOSLON grade	Number of cases
Grade I	3 (4.1%)
Grade II	18 (24.3%)
Grade III	22 (29.7%)
Grade IV	27 (36.5%)
Grade V	4 (5.4%)

The sample comprised 40 males (54%) and 34 females (46%) ( $p = 0.038$ ). Fifty-three (71.6%) of UCLP cases were left-sided clefts; whereas, 21 (28.4%) of the cases were right-sided ( $p < 0.001$ ). Intraexaminer agreement were very good for both assessors (0.89 for A and 0.83 for B) while the inter-examiner agreement was 0.74 showing good agreement between both assessors.

Table 2 shows dental arch relationships of Saudi UCLP children assessed by GOSLON Yardstick. Three children (4.1%) had excellent surgical outcomes (grade I), 18 children (24.3%) filled into grade II (good outcome), 22 subjects (29.7%) had grade III (fair outcome), 27 children (36.5%) had grade IV (poor outcome), and 4 subjects (5.4%) were ranked as having very poor outcomes (grade V). The mean GOSLON score was 3.39.

## DISCUSSION

Normal dental arch relationship is essential for proper speech, chewing, and swallowing. Dental malocclusion accompanied by oral clefting can make it challenging to maintain good oral health status. Thus, the child's confidence and their quality of life may be reduced. Therefore, early assessment and intervention should be encouraged. In this study, the GOSLON Yardstick was applied to categorize dental arch relationships of Saudi UCLP children. It has been proven to be the most accepted by the profession for assessing cleft deformities with a high level of sensitivity and reliability in comparing different surgical approaches. The Yardstick has shown the ability to detect the quality of dental arch relationships during all stages of dental development,<sup>14</sup> and predict surgical outcomes as early as 5 years of age.<sup>15</sup> However, the GOSLON Yardstick suffers certain deficiencies; the inherent subjectivity of the Yardstick and its inability to predict growth patterns in patients born with UCLP limit its use in all cleft deformities.<sup>16</sup> In addition, the predictive validity of the GOSLON Yardstick was influenced by gender where males showed worse deterioration in scores than females.<sup>17</sup>

A higher distribution of left-sided clefts (71.6%) was seen within the Saudi sample ( $p < 0.001$ ). This matches previous findings of orofacial clefts.<sup>18,19</sup> The predominance of left-sided cleft is a common finding in most cleft publications, but the etiology responsible for this predilection is still unclear.<sup>20</sup> In addition, the gender distribution of the Saudi UCLP sample showed that males had a higher incidence of this particular cleft deformity (54%,  $p = 0.038$ ) which is in accordance with literature on Caucasian populations.<sup>21,22</sup>

In this study, the age range of 8–10 years was chosen to avoid the effect of orthodontic treatment as a confounding variable and

to allow for cross-comparisons between studies. The careful analysis and comparison of current results with other cleft centers revealed that treatment outcomes based on dental arch relationships were inferior to most cleft publications (mean GOSLON score, 3.39). At the same age range (8–10 years), the GOSLON scores were 2.10 and 2.58 for Goteborg and Stockholm Sweden samples, respectively.<sup>23,24</sup> The GOSLON Yardstick score was 2.29 in Austria,<sup>25</sup> 2.36 in Nijmegen/Netherlands,<sup>26</sup> 2.44 in Warsaw/Poland,<sup>27</sup> 2.75 in Paraiba/Brazil,<sup>28</sup> 2.85 in Fukuoka/Japan, and 3.15 for Malay UCLP children.<sup>29</sup> Only few studies reported higher GOSLON scores than the current study; the New Zealand sample had 3.50 GOSLON score,<sup>30</sup> 3.50 in Tokyo sample/Japan,<sup>31</sup> 3.53 in Bristol/UK,<sup>32</sup> while the American cleft sample (Americleft B) had the worst surgical outcomes with GOSLON score of 3.66.<sup>33</sup>

The type and timing of lip and palate repair are important factors that affect the surgical outcome in UCLP children. In 1958, Dr. Millard introduced his rotational advancement method in lip repair. This was a groundbreaking chapter in the history of cleft lip care. Interestingly, Sitzman et al. reported that 84% of surgeons in the United States and Canada still perform rotation advancement techniques for complete unilateral cleft lip.<sup>34</sup> However, the Millard technique suffers several deficiencies. Millard's method seems to be less efficient when tissue deficiency is marked (complete unilateral cleft lips). This may explain why the lengthening of both the medial and lateral labial segments is compromised with Millard's repair.<sup>35</sup> Moreover, a greater frequency of hypertrophic scars following rotation-advancement repairs was reported in most cleft publications. This may increase the risk of developing negative anteroposterior and transverse dental arch relationships. On the contrary, the timing of hard palate closure, rather than the type of closure or sequence of hard or soft palate repair, determined the postoperative craniofacial complex growth. More growth inhibition was seen with earlier palate repair. The results from the Zurich and Goteborg centers, where the hard palate closure was performed at 7 and 9 years of age, respectively, had shown satisfactory facial growth.<sup>14,36</sup> On the contrary, palate closure at 18 months resulted in more sagittal and vertical growth inhibition as well as a reduction in posterior facial height.<sup>37</sup>

It should be noted that no nasoalveolar molding (NAM) was performed in the study sample; NAM is a presurgical infant orthopedics that involves alignment and approximation of the alveolar processes before lip repair. The procedure helped surgeons make the primary repair of the lip, alveolus, and the nose an effortless procedure. Studies have shown that NAM reduced significantly the need for surgical revisions and excessive scar tissue formation.<sup>38</sup> In addition, later alveolar bone grafting achieved better graft site stabilization and alveolar ridge contour after NAM fitting.<sup>39</sup>

The major limitation of this study is the assessment of dental arch relationships in cleft patients relying solely on a particular surgical protocol while ignoring other approaches applied by other surgeons which may have significant limitations and repercussions on treatment outcome analysis. By fixating on a single surgical protocol, the full spectrum of surgical techniques and their potential impact on dental arch relationships may be overlooked.

A more holistic approach, considering multiple surgical protocols, is essential to capture the nuanced and varied aspects of dental arch development in cleft patients, ultimately contributing to more effective treatment plans. In addition, Further studies that measure the effect of cleft surgery on soft tissue volumes and their relations with dental arch relationships may help develop

predictive therapeutic models with the aid of recent 3D imaging and computer-assisted technologies.

## CONCLUSION

The UCLP Saudi sample has a higher incidence of left-sided clefts and male tendency. Based on the dental arch relationships, the treatment outcome of UCLP Saudi children was unsatisfactory, with a mean GOSLON score of 3.39. Delayed palate repair and the use of presurgical orthopedics may be considered in the future for the management of cleft deformity. The GOSLON Yardstick can assess dental arch relationships among children with UCLP and further constitute a basis to evaluate favorable or unfavorable maxillary growth related to various treatment protocols.

## Clinical Significance

The current findings suggest that the cleft surgical protocol may be revised to improve treatment outcome in terms of dental arch relationships which is currently unsatisfactory.

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