Scholars Journal of Applied Medical Sciences
Abbreviated Key Title: Sch J App Med Sci
ISSN 2347-954X (Print) | ISSN 2320-6691 (Online)
Journal homepage: www.saspublishers.com

OPEN ACCESS

Outpatient Clinic

Skeletal Outcomes Assessment of Unilateral Cleft Lip and Palate Repair in Tunisia- About a Comparative Study

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DOI: 10.36347/sjams.2019.v07i12.037 | **Received:** 14.12.2018 | **Accepted:** 21.12.2018 | **Published:** 26.12.2019

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Abstract **Original Research Article**

***Introduction*:** Up to now, various techniques and protocols have been proposed for the repair of complete unilateral cleft lip. An objective assessment of the surgical techniques used and the timing of those procedures is imperative to evaluate the efficiency of each treatment modality. ***Materials & Methods*:** we conducted a comparative retrospective study in which we used both clinical and radiographic findings of 60 patients treated at the orthodontic department of the dental faculty-Monastir. To assess the therapeutic outcomes of unilateral cleft lip and palate repair, we used a cephalometric analysis for the skeletal outcomes. ***Results*:** Comparing the results of the groups among the different departments participating in the study showed better outcomes in maxillofacial surgery department of Sahloul significant higher mean maxillary prominence and larger convexity. However, no statistically significant differences were found for any of the mandibular measurements. ***Conclusions*:** According to our study, better craniofacial form can be due to a better surgical protocol followed when treating patients with unilateral clefts.

Keywords: Skeletal, clefts, unilateral, assessment, comparison, multicentric.

INTRODUCTION

For many years in the past, the array of surgical treatment approaches and primary infant management protocols for patients with cleft lip and palate was enormous, with little reliable information available upon which to base rational decision-making in choosing one method over another.

Up to now, many various techniques and protocols have been proposed for the repair of complete unilateral cleft lip [1].

For instance, an objective assessment of the surgical techniques employed to repair clefts of the lip and palate, and the timing of those procedures, is imperative to evaluate the efficiency of each treatment modality and aid the development of evidence-based guidelines for cleft lip and palate care.

Facial growth and skeletal relationships are of the key areas of interest for assessing and comparing the quality of cleft treatment outcome.

The main objective of this study, conducted in the orthodontic department in the dental faculty of Monastir, was to describe and compare the craniofacial morphology of patients with repaired complete unilateral cleft lip and palate surgically treated in different departments.

MATERIAL AND METHODS

The subjects of this retrospective longitudinal cohort study were obtained from the orthodontic department of Monastir- Tunisia and referred from four Tunisian departments: Maxilo-facial surgery department of Sahloul-Sousse, Pediatric surgey department of Monastir, Maxillo-facial surgery department of Sfax and Maxillo-facial surgery department of Rabta-Tunis.

Consecutively treated Tunisian individuals with a history of non-syndromic complete unilateral cleft lip and palate matched for age and sex from each participating department were selected. One of the challenges in conducting this kind of research is that clefts of the lip and palate present with great heterogeneity. The only substantial category that is reasonably homogenous is non-syndromic complete unilateral cleft hence its selection for this investigation [2].

The main considerations when establishing the methodology for this investigation were:

Inclusion criteria

- Unilateral cleft lip and palate: For most outcomes, samples must be separated according to cleft type. Since unilateral cleft lip and palate represents the most common cleft type, this cleft type was chosen for this study. Due to the variability in presentation in incomplete clefts, only patients with complete unilateral cleft lip and palate were included to keep the samples homogeneous and enabling comparability.
- Tunisian subjects: Since craniofacial form varies by race, to avoid the introduction of additional bias only Tunisian subjects were included in this investigation.
- Availability of records: Records had to be available to document and confirm the patient condition (reports, radiographs and study models). Records describing the surgical treatment protocol had to be available for each patient. Furthermore, all patients had to have received all of their primary surgical repairs at the respective department.
- Patient's acceptance: The patient permission had to be necessarily obtained to use their records in outcomes assessment.

Exclusion / Non-inclusion criteria

- Syndromes: Any patients with associated anomalies or syndromes were excluded.
- Antecedent of treatment: Any patients who received orthodontic treatment/intervention (fixed or removable appliance), headgear, or face-mask therapy, who underwent orthognathic surgery or osteogenesis distraction prior to the date of acquisition of the cephalometric radiograph selected for inclusion in this study were excluded.
- Lack of findings: incomplete or missing data within the medical record.

Referring to the original Eurocleft study and Americleft study, the following descriptive data were recorded for each subject included in this investigation. These data are in relation with:

Patient

- Date of birth/ Age of patient
- Sex of patient
- Cleft aspect : Type /Side of the cleft
- Overall, the sample comprised 60 individuals with an average age of 16 years, 10 months. The group consisted of 28 males (46, 66%) and 32 females (53, 33%).

The cleft was located on the left side in 37 patients (61, 66%) and on the right side in 23 patients (38, 33%).

The demographic characteristics of the sample are shown in Table I.

Table-I : Sample demographics

Department	N	MIF Ratio	Mean Age	Age Range
MFS Sahloul	23	10/13	17 yrs 4 mo.	10 yrs 0 mo. - 23 yrs 6 mo.
MFS Sfax	6	2/4	16 yrs 2 mo.	11 yrs 11 mo.- 20yrs 8 mo.
PS Monastir	21	12/9	14 yrs 11mo.	11 yrs 4 mo. - 18 yrs 6 mo.
MFS Tunis	10	4/6	18 yrs 1 mo.	9 yrs 2 mo. - 24 yrs 0 mo.
Totals:	60	28/32	16 yrs 10 mo.	

Despite the relative homogeneity of this cohort, considerable between-case variation still exists, requiring reasonably large samples for statistical comparison. Consequently, a minimum sample of between 20 and 30 patients per treatment center was deemed essential for such comparison. Unfortunately, for some of the subsamples (i.e. maxillofacial surgery departments of Sfax and Charles Nicole-Tunis), it was impossible to collect a sample of such size .That is why we limit our study in a tow-departments-comparison.

Surgical management

A thorough description of each surgical treatment protocol was recorded, including:

- Department/Hospital where surgeries were performed.
- The technique/type and approximate age of lip, hard and soft palate repair.
- The technique/approximate age at which alveolar bone grafting was performed.

The treatment protocols of the participating departments for patients with unilateral cleft lip and palate are described below and summarized in Table II.

Table-II: Summary of participating departments and their surgical treatment protocols.

Treatment	PS Monastir	MFS Sahloul
Cheiloplasty	3 mo. Malek technique ++	6 mo. Modified Millard technique
Veloplasty	6 mo. Depends on the case	6 mo. Three-plan-suture (Palatal fibromucosa +velar muscle+palate)
Uranoraphy	18 mo. Palatal-nasal mucosa suture	18 mo. Palatal-nasal mucosa suture
Alveoloplasty	3 y.	Primary: 4 to 5 y. Secondary: Alveolar cleft : Gingivoperioplasty Wide alveolar cleft: Alveolar graft
Rhinoplasty	No (except functional rhinoplasty)	4 to 5 y.
Secondary surgeries **Orthognathic surgery**	After completion of growth	After completion of growth

To achieve our research goals, we used both clinical and radiographic findings. A lateral cephalometric radiograph, furnished for each subject, was required for this investigation. In most instances, these radiographs were taken prior to the secondary alveolar bone grafting procedure. The radiographs were produced in conventional film format using the cephalometric equipment available, and acquired in either a hardcopy or digital format. For those lateral cephalograms obtained in conventional film format, an Epson Expression scanner was used to convert the hardcopy radiographs (analog image) into JPEG format (digital image).

These lateral cephalograms, now obtained or directly received in a digital file format were imported into the cephalometric software and their resolution subsequently adjusted as necessary, to resize the radiographs to match the dimensions of the hardcopy films so that relative uniformity could be achieved.

Finally, since the lateral cephalograms were produced using various types of radiographic equipments, possessing different enlargement factors, an arbitrarily set linear distance basion to nasion (Ba-N) of 110 mm was used to size-adjust all linear measurements to control for radiographic magnification discrepancies (Figure 1) [3,4].

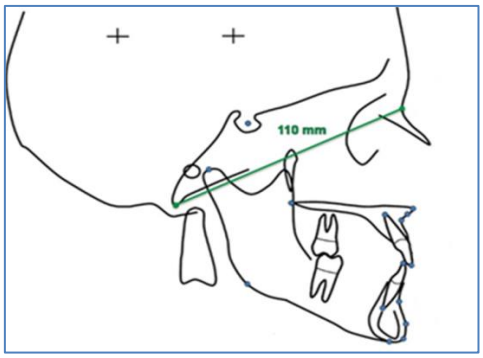

Fig-1: Cephalometric Size–Adjustment of the Cranial Base (Ba-N distance).
All cephalograms included in this investigation were scaled to an identical length
(Ba-N distance of 110mm)

The digital images were visually enhanced on the computer monitor using magnification, brightness, and contrast adjustments.

Autodesk AUTOCAD 2016 Software version was used for all cephalometric analysis. It was necessary to digitally plot 13 landmarks per radiograph to generate a cephalometric tracing (note: bilateral structures were mid-planed). Of these, 4 maxillary and 6 mandibular cephalometric landmarks were selected for inclusion in this investigation (Figure 2) (Table III).

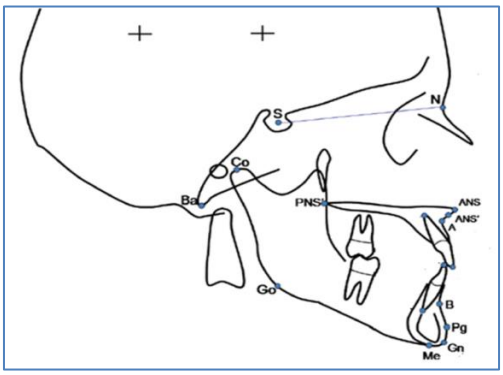

Fig-2: landmarks per radiograph to generate a cephalometric tracing

Table-III: Hard-Tissue landmarks

Hard-Tissue Landmarks	
A-point	The deepest point on the anterior contour of the maxillary alveolar process
ANS	The tip of the bony anterior nasal spine at the inferior margin of the piriform aperature, in the midsagittal plane
ANS'	The deepest point on the concavity of the anterior surface of the maxilla in the midline, within 3 mm of the floor of the nose
B-point	The deepest point on the anterior contour of the mandibular alveolar process
Ba	Basion: the most anteroinferior point on the margin of the foramen magnum
Co	Condylion: the most posterosuperior point on the head of the mandibular condyle
Gn	Gnathion: the most anteroinferior point on the outline of the chin
Go	Gonion: the most posteroinferior point on the angle of the mandible
Me	Menton: the most inferior point on the outline of the chin
N	Nasion: the most anterior point of the frontonasal suture
Pg	Pogonion: the most anterior point on the outline of the chin
PNS'	Point of intersection of a vertical line projected down from PTM (perpendicular to a plane constructed 7˚ up from SN) and the palatal plane
S	Sella: the geometric center of sella turcica

Utilizing these landmarks, 15 hard-tissue cephalometric measurements were evaluated: 8 angular, 6 linear, and 1 ratio 5.

The measurements were based on those employed in the original Eurocleft study with some modifications (Table IV).

Table IV: Hard-tissue cephalometric measurements assessed.

Hard-Tissue Cephalometric Measurements
Ba-N (mm)
SNA (˚)
SNB (˚)
ANB (˚)
Ba-N-ANS' (˚)
Ba-N-Pg (˚)
ANS'-N-Pg (˚)
WITS appraisal (A†OP: B†OP) (mm)
Md length (Co-Gn) (mm)
SN-MP (SN-GoGn) (˚)
AUFH (N-ANS') (mm)
ALFH (ANS'-Me) (mm)
ATFH (N-Me) (mm)
Lower face height ratio (ANS'-Me/N-Me) (%)
Md Axis angle (BaN-CoGn) (˚)

RESULTS

This study showed good results with procedures and techniques followed in Maxillo-facial surgery department of Sahloul. However, there are significant differences in the skeletal measurements among unilateral cleft patients of the two departments.

The most interesting finding was the significantly higher mean of maxillary prominence in maxillo-facial surgery department of Sahloul (SNA of 79.8°; Ba-N-ANS' of 62.7°). Pediatric surgery department of Monastir had the lowest mean maxillary prominence (SNA of 76.3°; Ba-N-ANS' of 59.9°).

When assessing maxillo-mandibular relationship, maxilla-facial surgery department of Sahloul showed a significantly larger convexity (ANB of 3.4°; ANS'-N-Pg of 6.0°).

This department also exhibit the steepest mean mandibular plane angle and the longest anterior lower face height. However, no statistically significant differences were found for any of the mandibular measurements, although Sahloul's department tended to have the shortest mean mandibular length.

The results of the comparisons of the groups among the departments participating of the study are shown in Table V.

Table-V: Mean values for the hard-tissue measurements

Measurement	**PS Monastir**	**MFS Sahloul**
Ba-N (mm)±	110	110
SNA (˚)	76.30***	79.80***
SNB (˚)	75.90	76.38
ANB (˚)	0.40**	3.42**
Ba-N-ANS' (˚)	59.93**	62.77**
Ba-N-Pg (˚)	56.53	56.70
ANS'-N-Pg (˚)	3.39*	6.07*
WITS appraisal (mm)	-1.40	0.57
Md length (Co-Gn) (mm)	110.30	109.55
SN-MP (SN-GoGn) (˚)	33.98	36.51
AUFH (N-ANS') (mm)	52.60	52.40
ALFH (ANS'-Me) (mm)	64.04	65.76
ATFH (N-Me) (mm)	116.65	118.17
Lower face height ratio (ANS'-Me/N-Me) (%)	54.88	55.61
Md axis angle (BaN-CoGn) (˚)	75.76	76.40

Bold numbers are statistically significantly different from each other.

** p<0.05, ** p<0.01, *** p<0.001, denotes the level of significance in comparison.*

DISCUSSION

Numerous authors have emphasized the relative strengths of evidence obtained through various reporting methods and types of investigations: The strongest form of available evidence are represented by meta-analyses and randomized control trials which, admittedly for cleft lip and palate treatment, are extremely difficult to execute due to time, costs and the large samples required [5].

In the hierarchy of evidence, intercenter comparisons of outcomes are considered second strongest behind randomized control trials, assuming appropriate interpretation of results and rigorous attempts to control bias [6].

Regardless of study design, however, quantifying the effect of surgical management of unilateral clefts and the heterogeneity of the presenting condition remain a challenge.

Bias has been identified by many authors as a primary weakness that limits the strength of evidence from retrospective studies [6].

Nonetheless, the implementation of a well-designed intercenter comparison study provides an opportunity to control several of these biases, providing greater transparency and therefore increased integrity of the results versus single-center reports [2].

Intercenter studies such as Eurocleft and Americleft investigation contain the risk that any significant differences found in craniofacial form may have arisen not only from differences in the treatment protocol employed by each center but also because the populations compared were fundamentally dissimilar.

Specifically, ethnic, racial, genetic and age differences all represent sources of variability that could contribute to the differences observed [3].

These inherent differences among the participating centers patient population reduce the validity of comparisons made between them when relating variations in craniofacial morphology and treatment protocol.

Therefore, to minimize the confounding effect of susceptibility bias on the outcomes measured, only treated non-syndromic unilateral cleft lip and palate Tunisian subjects of approximately the same age were included in this investigation.

In addition to the numerous potential research biases, one of the greatest obstacles to meaningful intercenter outcome comparisons is the inconsistency and non-comparability of the records that are taken to document those outcomes. This is usually due either to a lack of consistent protocols within the center for record-taking and a lack of oversight by the responsible team members for records to be taken. Therefore, the use of standard measurements and scores was critical to this investigation.

To describe skeletal outcomes, we used skeletal measurements and cephalometric radiographs, taken by the residents in the orthodontic department of Monastir as a routine part of treatment planning. This is necessary to reduce patient risk from additional X-ray exposure and in consideration of the cost of additional records.

In the original Eurocleft study, the need for international standardization of the specifications of radiologic cephalometric equipment was identified.

In an effort to circumvent the issue of variable radiographic enlargement factors in the present investigation, the technique utilized by Ross, of size-adjusting all linear distances to an internal standard (basion to nasion distance of 110mm) was employed [4].

Concerning the use of computers for cephalometric analysis, authors reported that computer use did not increase cephalometric measurement error when compared with hand tracing. More recently, Farman evaluated the reproducibility and accuracy of cephalometric landmark identification between indirect and direct digital images using the conventional image as the ‘gold standard.

The authors found that while landmark identification was statistically significantly more reproducible for hard- and dental- tissue landmark identification for a direct compared to indirect digital images, cephalometric measurement was equivalent in accuracy to conventional film.

According to the literature, in patients with repaired unilateral cleft lip and palate, some skeletal deformities of the maxilla are seen to occur frequently:

- Disturbance in downward growth of the maxilla: Following surgical lip and palate repair, authors describe vertical insufficiency of the maxillary jaw associated with alveolar processes and palate [7, 8].
- Retrognathism and reduction of transverse growth of the maxilla: Brachymaxillia
- Medial collapse of the upper alveolar arch: As same as transverse reduction of the maxilla, collapse of the alveolar arch has been related to the muco-periosteal flap repair of the palate, while some other authors accused the tightness of the upper lip, result of lip repair, to be responsible for this collapse of the upper alveolar arch [7].

Studies on untreated adult patients with unilateral cleft lip and palate showed a smaller mandibular body and ramus height, a tendency for the

mandible to be retruded, and an increased mandibular plane angle in patients with clefts, compared with noncleft subjects [9].

An overview of literature provides several theories attempting to explain the inferior-posterior mandibular inclination in patients with cleft with involvement of the palate. A functional explanation could be logic, with nasal obstruction causing mouth breathing. However, Molsted et al. Suggested that both passive and active presurgical orthopedics alter the tongue position and causes the inferior and posterior mandibular inclination.

Other investigators found a significant correlation between the degree of posterior mandibular rotation and the degree of maxillary retrusion in unilateral cleft- patients and therefore explained the positional changes of the mandible as a compensation mechanism for maxillary retrusion [10].

While da Silva reported that different surgical protocols had no influence on mandibular characteristics and confirmed his hypothesis that mandibular morphology and growth direction are inherent to the cleft and are not vulnerable to surgical procedures, recent studies showed significant less posterior vertical maxillary development, a more open maxillary plane, and a tendency to a more retruded mandible as cleft surgeries side-effects [11].

Not only surgical intervention does interfere with growth of the skeletal component of the face because of the scar tissue in lip and palate having a restraining effect on growth, but also genetic inheritance, cleft type and size initially determine the patients Craniofacial morphology. Data suggest that in patients with complete unilateral cleft, there is a significant relationship between initial cleft severity and maxillary growth: In fact, Patients with a small cleft area have a more protruded maxilla than do those with a large cleft area [7].

In addition, there has been some assumptions that the cleft malformation itself may explain the maxillary deformities, quite apart from the surgeries undergone to repair lip and palate: Walker has supposed that agenesis associated with the cleft may be an important factor in reducing the maxillary growth [7].

Strength and weakness of the study

The significance of this investigation lies in its potential value in the quest for information that would help cleft lip and palate teams identify which treatment protocols produce the most favorable outcomes in terms of craniofacial appearance. The findings generated are meant to prompt teams to perform clinical audits providing them with a basis against which to evaluate their own established treatment protocols for potential modifications.

Since most departments rendering clefts care employ their own specific treatment protocol, and given the overwhelming number of different approaches used, it is incumbent upon those treating patients with to compare and scrutinize their outcomes methodically and scientifically in order to make evidence-based decisions regarding treatment choices offered to patients. Thus, collaboration between those departments is essential [12, 13].

Such collaboration, as started in Europe in the late 1980's, has led to a rapid growth in our knowledge base, has generated standards for recording and reporting outcomes, and has laid the groundwork for more sophisticated investigations, namely, the prospective randomized controlled clinical trials presently being conducted by a collaborative group in Scandinavia, known as Scandcleft [2].

Specefically in Tunisia, While the large number of departments or centers and individuals providing treatment for craniofacial anomalies improves patients' geographical accessibility to care, it simultaneously creates a fractionation of the study population thereby reducing the probability of developing patient samples of adequate size to enable valid research.

The entire landscape is further complicated by non-comparable patient populations, non-comparable treatment records, unquantifiable differences in operator skills, and difficulties in letting go of biases.

CONCLUSION

According to our study, significant differences in craniofacial morphology exist among patients with unilateral cleft lip and palate treated following different surgical protocols at the departments investigated in this study.

No significant differences in mandibular prominence or vertical dimension were found among the patients participating in this investigation.

In summary, one of most valuable outcomes of this study has been the experience and insight gained in understanding the requirements, demands, and possible obstacles that must be overcome. It is my hope that the present investigation, despite its several confounding factors, represents a first step toward a more generalized collaboration among Tunisian departments, agreement on standardized record-taking to allow comparability, and the eventual organization and execution of prospective randomized controlled clinical trials on large numbers of patients, to enable the identification of "good practice" protocols.

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