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Anatomy-imaging Analysis of the Labio-Maxillo-Palatine Clefts with Getting Printed 3D Models

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Abstract

Labio-maxillo-palatine clefts are malformations of viscerocranium that require a multidisciplinary team for establishing a diagnosis and treatment, involving multiple specialties such as plastic surgery, maxillofacial surgery, ENT, paediatrics, speech therapy, orthodontics. For the accuracy of the diagnosis and treatment these patients require multiple explorations that will involve multiple doses of irradiation. The exploration of these patients starts at an early age and that's why should have a limited indication. CBCT is a modern exploration that offers a low dose of irradiation. The principle that CBCT uses is similar to the principle of explorations real-time imaging in operating rooms when surgery takes place. Bone defects in the labio-maxillo-palatine clefts require a correct evaluation for the planning of the augmentation intervention of these defects, and the CBCT allows for an optimal 3D assessment to determine their dimensions and the bone required. The use of these explorations facilitates the operating steps. With the development of CBCT, visualization programs and analysis of these explorations were also developed, with necessary tools in linear and volumetric measurement of alveolar bone defect. We performed a retrospective study on patients with labio-maxillo-palatine clefts, diagnosed in the Paediatric Surgery Clinic, Saint Mary's Children's Hospital, Iasi in 2019 to demonstrate the need for an accurate diagnosis using this radiological technique, CBCT and digital planning, and on printed 3D models of surgical interventions to correct alveolar bone defects of the labio-maxillo-palatine clefts.

Keywords: Labio-Maxillo-Palatine clefts • CBCT • Alveolar Bone Defects • 3D Exploration

Introduction

The development of congenital facial abnormalities of the lips and palate usually takes place between the fourth and twelve weeks of intrauterine life due to a lack of fusion of jaws and palatal apophasis [1]. Various imaging methods have been used to evaluate labio-maxillo cleavage and to follow bone-graft treatments [2]. When these examinations are carried out before bone grafting surgery, they allow to estimate the size, position and structures involved in the labio-maxillo-palatine cleft Computed tomography (CT) allows precise evaluation of the shape, quality (spongy and cortical), height and thickness of the bone by using reconstitutions on several levels [3]. The use of CT, using three-dimensional (3D) protocols, allows excellent visualization of bone architecture and is considered a valuable tool in the evaluation of cranio-facial deformity in patients with congenital malformations, such as labio-maxillo-palatine cleft [4]. There are well-defined protocols for the treatment of labio-maxillo-palatine clefts from the prenatal period to adulthood, but there are no studies related to the development of a methodology capable of determining, in volumetric imaging examinations, the volumetric size of the bone defect. CBCT is a new technology in the field of oral and maxillofacial imaging and allows a three-dimensional visualization of the scanned anatomical regions. In the specialized literature, CBCT was also called dental tomography computer, volumetric tomography computer or volumetric tomography. The term Cone Beam CT seems to be preferred by most practitioners because it best describes the principle of operation. The CBCT allows a volume scan using a conical beam of X-rays, thereby reducing the exposure time and irradiation dose.

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Considering that patients with labio-maxillo-palatine clefts should be operated on from an early age, this method has the advantage of a considerably reduced irradiation dose compared to conventional CT used in current medical practice. In the decision to repair the deformities of the cleft of the lip and palate, the development of speech, facial growth, the psychological effects of the child, and the family, and the safety of anaesthesia are taken into account. In patients with cleft palate with or without cleft of the lip, the placement of the tympanostomy tube is performed from the age of 3 months to 6 months. This operation is performed at an early age to minimize the likelihood of a chronic ear disease and loss of the auditory duct due to aeration of the middle ear. In patients with cleft palate and lip, the tympastomy tube is placed simultaneously to repair the cleft of the lip. Repair of the cleft palate is carried out before the initiation of speech development. This is usually carried out from nine to 15 months. The long-lasting action of the tympanostomy tube is put during palatoplasty. The craniofacial and cleft team closely monitors speech development after repairing the cleft palate from the age of two. Velopharyngeal dysfunction (VPD) should be evaluated with nasopharyngeoscopy and fluoroscopy video. If the degree of VPD is signifying, speech therapy is undertaken at about three years. If the aggressive logopady is unsatisfactory, the surgical correction of the VPD must be performed between four and six years. Subsequent surgical treatments for cleft deformities include grafting of the alveolar bone from the age of 9 years to 11 years, external and internal nasal reconstruction (cleft of septorinoplasty) at the age of 12-18 years and orthognant surgical operation after the development of the facial skeleton is complete. The need for these interventions is related to the initial deformation, as well as the development of the patient's growth.

The objectives of this work are:

- Making linear measurements of the alveolar bone defect that were obtained by exploring CBCT in patients with labio-maxillo-palatine cleft using Romexis as software.
- Making linear measurements of the alveolar bone defect that were obtained by exploring CBCT in patients with labio-maxillo-palatine cleft using radiAnt as software.
- Making linear measurements of the alveolar bone defect in patients with labio-maxillo-palatine cleft using printed 3D models resulting from cbct exploration.

- Comparing the measurements obtained by the two software's Romexis and RadiAnt with printed 3D models.

Materials and Methods

The study was conducted to analyse the linear dimensions of the alveolar bone defect present in patients with labio-maxillo-palatine clefts using a three-dimensional imaging exploration, namely cone beam CT (CBCT) with two analysis and visualization programs and printed 3D models following the realization of CBCT exploration.

Selection of patients in study groups

In this study, 7 patients were selected who presented themselves in the paediatric surgery clinic, Saint Mary's Children's Hospital, Iasi for the surgical treatment of labio-maxillo-palatine clefts and who were examined with the help of CBCT in 2019.

The exclusion criteria were:

1. A treatment history for oral breathing during sleep, including tonsillectomy, adenoidectomy or recurrent tonsillitis
2. Frequent colds (6 or more per year)
3. A history of dysphagia and continuous positive airway pressure therapy.

Another exclusion criterion for control groups was the existence of any type of syndrome. All witness subjects had a normal craniofacial morphology without jaw deformities. Patients had an average age of 7.85 years and all patients had a unilateral localization of labio-maxillo-palatine cleft. The closure of the defects by the Millard-type surgical technique was performed in most patients, and the closure of the palate was performed by palatoplasty. None of the patients with DLMP underwent any maxillary expansion surgery or alveolar bone grafting at the time of inclusion in the study.

CBCT exam protocol

Prior to the CBCT scan, the parents of the patients were fully informed about the purpose of this study and the risks associated with CBCT. The CBCT equipment used was planmeca Promax 3D Mid (Planmeca OY, Helsinki, Finland). The scan was performed by selecting an FOV and the following exposure parameters: kV, mA, seconds and voxel size depending on the patient's age and weight (Tables 1 and 2) the initial and final reconstructions were carried out by the software Romexis 3.6.0 (Planmeca, Helsinki, Finland). In order to make axial, sagittal and coronal sections, CBCT reconstructions with a thickness of 1 mm and at a distance of 1 mm were established.

Each patient was seated on a chair with the Frankfurt plane parallel to the floor, asked not to move his head or swallow, to keep the arches in the position of centric occlusion, with his tongue and lips relaxed, during the examination.

Linear analysis of alveolar bone defect on CBCT analysis programs

The digital images obtained were transferred directly from the CT scanner to a personal computer for image processing and storage in a special format (DICOM). A CBCT technician identified the files, removed the name, gender and date of birth from DICOM. All the original axial sections were stored on CD-ROM in DICOM (Digital Imaging Communications in Medicine) and then transported to an independent workstation (Dell 1420 - Windows 10) containing the Romexis 4.0.1 program (Planmeca, Helsinki, Finland).

All CBTCs were independently analysed by an examiner – General medical student in the final year

Before analysing the images, the criterion used to determine the limits of the bone defect was established. The criterion used for the skull under examination was to trace the contour of the bone to the limit of the defect located at the level of the alveolar process of the maxilla. The images resulting from the exploration of CBCT were analysed using two programs - Romexis and Radiant. The instrument used in romexis 4.0.1 and radiant program allowed linear tracing with the measurement of the distance between the ends of the alveolar defect, using axial sections, thus measuring the transverse distance of the defect and the antero-posterior distance.

Linear analysis of alveolar bone defect on printed 3D models

After the CBCT exploration, the volume obtained in DICOM format was transferred to a 3D printer (3D Form 2 Form Labs) and thus it was possible to print a model of the skull with the bone defect but also with the perioral soft parts of each patient with labio-maxillo-palatine cleft. Before analysing 3D models, the criterion used to determine the limits of the bone defect was established. The criterion used for the printed skull was to trace the contour of the bone to the limit of the defect located at the level of the alveolar process of the maxilla. With the help of a linear meter, linear tracing was allowed with the measurement of the distance between the extremities of the alveolar defect, thus measuring the transverse distance of the defect and the antero-posterior distance (Figure 1).

Table 1. general characteristic of patients with DLMP.

Pacients	Age (years)	Sex	Cleft - location
1	6	M	Left
2	6	M	Right
3	7	M	Right
4	7	F	Right
5	10	M	Left
6	10	F	Right
7	9	M	Right

Table 2. Parameters of exposure in patients with DLMP.

Pacients - DLMP	FOV	kV	mA	s	DAP (mGyx cm 3)	Size voxel
1	20 × 17 cm	90	6.3	27	1570	400 × 400
2	20 × 17 cm	90	9	27	2242	400 × 400
3	20 × 17 cm	90	6.3	27	1570	400 × 400
4	20 × 17 cm	90	8	27	1992	400 × 400
5	20 × 17 cm	90	12	27	1570	400 × 400
6	20 × 17 cm	90	6.3	27	1570	400 × 400
7	20 × 17 cm	90	8	27	1992	400 × 400

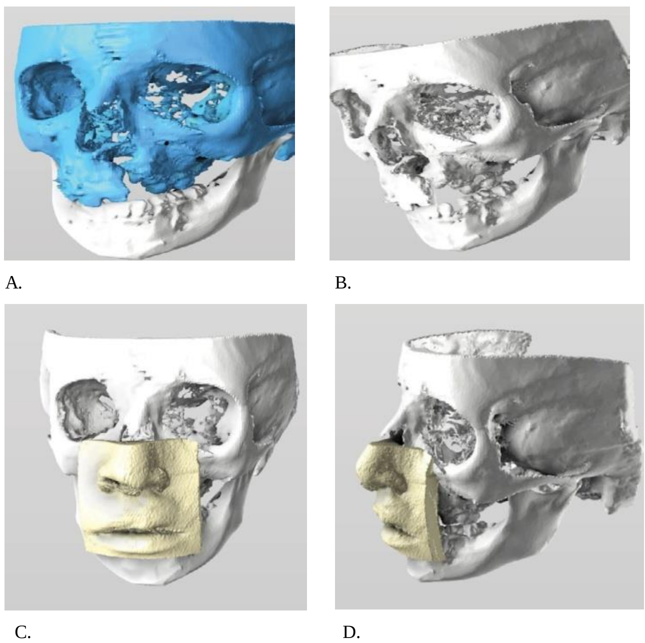

Figure 1. Graphic representation of the 3D models to be printed in a patient with labio-maxillo-palatine cleft-bone structure (A and B) and overlapping of the soft parts of the nasal and labial region (C and D).

Statistical analysis

In this research, the Microsoft Office EXCEL program and SPSS 20, dedicated to medical research, were used for statistical data processing. In the study, specific tests were applied to various types of analysed data, among which we can mention tests comparing the average values of a parameter corresponding to several batches of data, of which the ANOVA, Scheffé, Spjotvol/Stoline test, specific correlation tests for quantitative variables as well as for qualitative variables of which we can mention Pearson, Chi – square, Mantel-Haenszel, Fisher, Spearman, Kendall tau, Gamma. Following the application of these tests, the main parameters of interest were discussed and, depending on their values, the conclusions were established. Thus p the reference parameter calculated in the tests represents the level of significance of the test, which was compared with p=0.05 corresponding to a 95% confidence, which has significant values for the p calculate <0.05.

Results

As for the transverse distance analyzed in the Romexis program, the minimum value was 6.54 mm, and the maximum value was 12.6 mm, with an average value of 10.09 mm. regarding the transverse distance analyzed in the Radiant program, the minimum value was 5.56 mm, and the maximum value was 13.54 mm, with an average value of 10.33 mm. The analysis on printed 3D models regarding the transverse distance revealed a minimum value of 6 mm, and the maximum value of 12 mm, with an average value of 9.71 mm (Table 3). The results of the transverse distance for each evaluation technique and each patient correlated statistically significantly, as seen in the (Figure 2). As regards the antero-rear distance analysed in the Romexis programme, the minimum value was 4.07 mm and the maximum value 11.7 mm, with an average value of 7.48 mm. regarding the antero-rear distance analyzed in the Radiant program, the minimum value was 7 mm and the maximum value was 8.94 mm, with an average value of 8.10 mm. The analysis on the printed 3D models regarding the antero-rear distance revealed a minimum value of 7 mm, and the maximum value of 11 mm, with an average value of 8.14 mm (Table 4) (Figures 3 and 4).

Discussion

In the United States of America, 6800 labio-maxillo-palatine clefts (DLMP) are diagnosed annually, thus becoming the most common congenital malformation [5]. DLMP increases the risk of anatomical complications due to nasal abnormalities (septum deviation, narine atresia, hypertrophy of the nasal turbinate's, maxillary constriction [6]. These anomalies are largely attributed to the congenital defect itself and to surgeries performed to repair the orofacial defect [7]. Investigations such as profile and antero-posterior teleradiography, two-dimensional (2D) X-rays, plethysmography and rhinomanometry help to estimate the permeability's of the nasal [8]. A rhinomanometric analysis indicated that patients with bilateral DLMP show a 41% decrease in the nasal airway, and in those with unilateral DLMP a reduction of only 19 [9]. In the case of comparison between people with labio-maxillo-palatine clefts and those without plethysmography shows a 30% reduction [10]. Labial, palatal cleft, or compound forms are the most common congenital facial deformities, with an incidence rate of 0.65% among newborns, with ethnic and geographical variations [11]. Typical labial manifestations and cleft palate include on the one hand a deficit in one half of the facial massif, represented by a retreated jaw, but also a contralateral deviation of the nasal fossa, with the deviation of the nasal septum and the defamation of the base of the nose Patients with unilateral clefts normally experience facial distortions and asymmetrical maxillary bone defects 1990. Even after primary surgery to correct the defect to improve the appearance, patients with unilateral DLMP experience a distinct facial asymmetry In the nasal-labial region, distortion of the nasal septum and deviation to the affected side have been reported Some authors have reported significant differences in the lower third of the face, particularly in the mandible, while others have provided conflicting data. The small size and retrograded position of the mandible in adolescent patients with DLMP compared to adolescent patients in the control group could reduce the volume of the pharyngeal airway. However, the groups of adolescents with DLMP and control

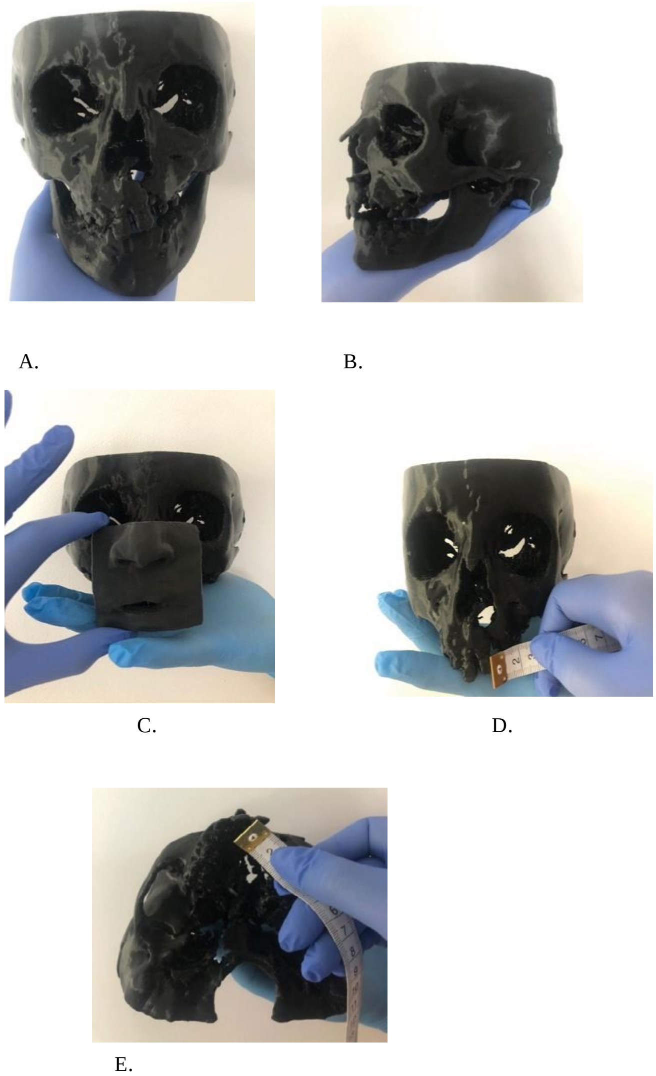

Figure 2. 3D printed models in patients with labio-maxillo-palatine clefts-face visualization (A), profile visualization (B), overlapping soft parts nasal and labial region (C), measurement of trasversal distance (D) and antero-posterior (E).

Table 3. Results regarding the transverse distance of the alveolar bone defect.

Transverse Distance-Romexis	Transverse Distance-Radiant	Distance on the Printed 3D Model
11.5	12.8	12
12.6	9.76	11
12.5	13.54	11
6.54	5.56	6
7.7	7	7
8.03	10.77	9
11.8	12.9	12

Table 4. Results regarding the antero-posterior distance of the alveolar bone defect.

Antero-posterior Distance-Romexis	Antero-posterior Distance-Radiant	Distance on the Printed 3D Model
11.7	7	11
6.7	8.53	8
8.14	7.55	8
6.73	7.94	7
4.07	8.48	7
6.03	8.3	7
9.01	8.94	9

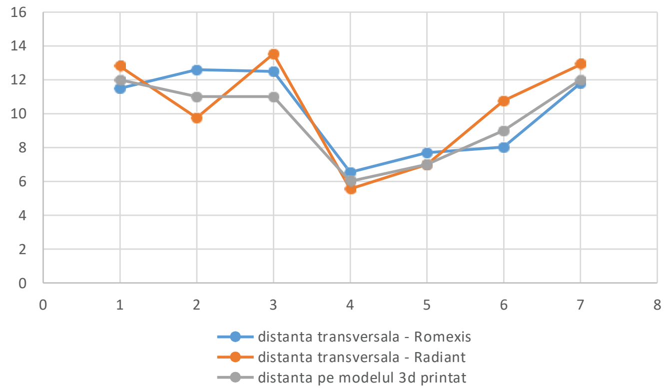

Figure 3. Graphical representation of the correlation between the three methods of measuring the transverse distance.

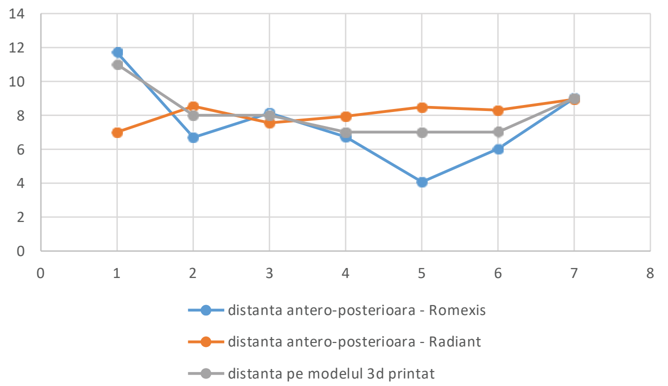

Figure 4. Graphic representation of the correlation between the three methods of measuring the antero-posterior distance.

showed no significant difference in the total volume of the pharyngeal airways. Moreover, although the volume of the nasopharynx in the adolescent DLMP group was significantly lower than that in the teen control group, the volume of the oropharynx in the adolescent DLMP group was significantly higher than that of the juvenile DLMP group. These dimensions support a compensatory phenomenon for increasing the volume of the pharyngeal airways in patients with DLMP. Nasopharyngeal insufficiency is a major problem in patients with DLMP, who have a narrowed volume of the pharyngeal airway compared to patients in the control group, as demonstrated in this study. In this context, the simple extension of the jaw and mandible could not be the best treatment option for the rehabilitation of respiratory function, because there is a risk that the nasopharyngeal insufficiency may be exacerbated. Further developments in surgical procedures and the change in growth through orthopedic treatments are necessary to fundamentally improve treatment options for patients with DLMP. The advantages of CBCT include a limited beam of X-rays, image accuracy, rapid scanning time, reduction of irradiation dose and reduction of artifacts. A number of recent studies have used CBCT explorations to assess airway in terms of facial morphology in people with sleep apnea [12]. The exhaustive search for the literature has so far not revealed any published CBCT studies on the airways of people with DLMP, despite the frequent respiratory problems that occur in this category of patients [13]. We hypothesized that people with labio-maxillo-palatine cleft have smaller pharyngeal areas and transverse volumes compared to the controls without DLMP. Profile tele radiographies of children with labio-maxillo-palatine clefts were compared with those of control patients without clefts, and the results were the reduction of bone nasopharyngeal frame and pharyngeal airways in the first category of patients. In addition, it was revealed a reduction of the upper airway in minors with labio-maxillo-palatine cleft, compared to children without cleft of the same sex and age, and this characteristic proved to persist during adolescence, when comparing profile tele radiographies [14].

Conclusion

Cbct exploration has become in recent years a basic technique in evaluating patients with skull malformations, such as labio-maxillo-palatine clefts. This study highlighted the need for digital planning of surgical interventions to correct defects encountered in labio-maxillo-palatine clefts, providing better surgical results when gathering more information related to preoperative preparation.

Acknowledgment

The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

Data Availability

The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

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