

## Surgical Premaxilla Reposition and Secondary Bone Grafting in a Children with Complete Bilateral Trans-Foramen Cleft Lip and Palate: a Case Report

*Reposicionamento Cirúrgico da Pré-maxila e Enxertia Óssea Secundária em uma Criança com Fissura Lábiopalatina Transforame Bilateral: Relato de caso*  
*Reposición Quirúrgica de la Premaxila e Injerto Óseo Secundario en Niños con Labio y Paladar Fisurados Transforamen Bilateral Completos: Reporte de un Caso*

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### Abstract

The objective of this study is to report the clinical case of a child with complete bilateral trans-foramen CLP who underwent surgical repositioning of the premaxilla with internal rigid fixation and a secondary bone grafting. The boy was following by different specialists since five days of life, being subjected to the entire surgical protocol to close the lip and palate. At 9 years old it was proposed surgical premaxilla reposition and, in a second surgery, a bone grafting in the alveolar process. After a virtual planning, surgery was performed under general anesthesia. The access was performed maintaining the vestibular pedicle of premaxilla, and after remove the interferences at the vomer and maxilla, premaxilla was fixed with titanium plates and screws. Six months after, fixation material was removed and the remaining gaps were grafted with iliac bone. After 21 months of the first surgery, the patient presents a good anteroposterior positioning of the maxilla and lip seal and, those responsible for the patient, reported an improvement in the patient's quality of life, with improvements mainly in speech, chewing and the patient's social life.

**Descriptors:** Cleft Lip; Cleft Palate; Osteotomy; Skeletal Fixation; Alveolar Cleft Grafting.

### Resumo

O objetivo deste estudo é relatar o caso clínico de uma criança com fissura lábio palatina transforame bilateral completa que foi submetida a reposicionamento cirúrgico da pré-maxila com fixação interna rígida e enxertia óssea secundária. O paciente foi acompanhado por diversos especialistas desde os cinco dias de vida, sendo submetido a todo o protocolo cirúrgico para fechamento de lábio e palato. Aos 9 anos foi proposta a reposição cirúrgica da pré-maxila e, numa segunda cirurgia, um enxerto ósseo no processo alveolar. Após planejamento virtual, a cirurgia foi realizada sob anestesia geral. O acesso foi realizado mantendo o pedículo vestibular da pré-maxila, e após retiradas as interferências no vômer e na maxila, a pré-maxila foi fixada com placas e parafusos de titânio. Seis meses após, o material de fixação foi removido e as lacunas restantes foram enxertadas com osso ilíaco. Após 21 meses da primeira cirurgia, o paciente apresenta um bom posicionamento anteroposterior da maxila e selamento labial e os responsáveis pelo paciente relataram melhora na qualidade de vida do paciente, com melhorias principalmente na fala, na mastigação e no social do paciente. vida.

**Descritores:** Fenda Labial; Fissura Palatina; Osteotomia; Fixação Óssea; Enxerto de Osso Alveolar.

### Resumen

El objetivo de este estudio es reportar el caso clínico de un niño con fisura transforamen bilateral completa de labio y paladar al que se le realizó reposicionamiento quirúrgico del premaxilar con fijación interna rígida e injerto óseo secundario. El paciente fue seguido por varios especialistas desde los cinco días de nacido, sometiéndose a todo el protocolo quirúrgico para cerrar el labio y el paladar. A los 9 años se propuso la sustitución quirúrgica del premaxilar y, en una segunda cirugía, un injerto óseo en la apófisis alveolar. Luego de una planificación virtual, la cirugía se realizó bajo anestesia general. El acceso se realizó manteniendo el pedículo vestibular del premaxilar, y luego de retirar la interferencia en el vômer y el maxilar, se fijó el premaxilar con placas y tornillos de titanio. Seis meses después, se retiró el material de fijación y los espacios restantes se injertaron con hueso ilíaco. A 21 meses de la primera cirugía, el paciente presenta una buena posición anteroposterior del maxilar y sellado labial y los responsables del paciente reportaron una mejoría en la calidad de vida del paciente, con mejoras principalmente en el habla, la masticación y el funcionamiento social del paciente. vida.

**Descritores:** Labio Leporino; Fissura del Paladar; Osteotomía; Fijación de Fractura; Injerto de Hueso Alveolar.

### INTRODUCTION

Cleft lip and palate (CLP) are congenital craniofacial malformations that represent a non-union of the upper lip and/or palate uni or bilateral that lead, in the severe cases, to a complex change in the maxillofacial growth and several dental and alveolar changes<sup>1</sup>. The most used classification of CLP was proposed by Sipina in 1972 that uses the position of the cleft in relation to the incisive

foramen, then could be: pre-foramen, trans-foramen and post-foramen<sup>2</sup>.

In Brazil, it is estimated that the prevalence of cleft children is 52 children per 100 000 live births<sup>3</sup>. CLP represents an important public health problem because for one side they are functional, aesthetic, and psychosocial implications for the patient<sup>4</sup> and on the other hand the treatment is complex and must need a multidisciplinary approach<sup>5</sup>.

This treatment starts in the newborn and several surgical procedures like cheiloplasty, palatoplasty, bone grafting and others are carried out involving different specialists<sup>5</sup>, and the complexity of treatment depends on the type of cleft<sup>6</sup>.

The treatment of bilateral trans-foramen CLP patients is the most complex one because of factors related to deficiency of soft tissue and bone evaluable to covering the cleft, the possibility of damage to the permanent dentition, the severe anterior projection and mobility of the premaxilla, and others<sup>8</sup>. Thus, surgical techniques are being created and discussed to help this surgical management of the premaxilla repositioning, such as developed by Aburezq<sup>9</sup> and modified by Carlini<sup>4,8</sup>. The most important difference between these techniques is the Carlini proposed an internal rigid fixation of the premaxilla after the repositioning.

The Carlini & Biron 2020 study showed that using this technique, they have success in 15 of 16 children in six months of follow-up<sup>8</sup>, that suggested a possible solution to the problems related to the premaxilla repositioning. However, this technique is new and is not a consensus in literature about the surgical management of the premaxilla.

The following case shows a premaxilla repositioning with internal rigid fixation, followed by a second bone graft with 21 months of follow-up in a boy with nine years old with complete bilateral trans-foramen CLP associated a severe projection of premaxilla.

### CLINICAL CASE

A boy with complete bilateral trans-foramen CLP was initially attended at five days of age at comprehensive care center for cleft patients of Curitiba, Brazil (CAIF). The family history showed a maternal uncle with cleft palate.

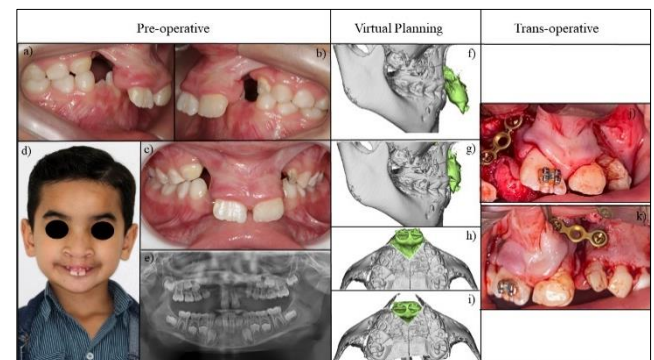
At 3 months of age a cheiloplasty associated with primary rhinoplasty was performed. Then, with one year and five months a complete primary palatoplasty was done and he was followed by a speech therapist. At five years old, the intraoral examinations showed a severe anteroinferior projection of the premaxilla (Figure 1 – d, e, f) causing loss of lip seal (Figure 1 – a, b). At this moment it was started the orthodontics treatment, a maxilla expansion was performed through a Hass apparatus. After expansion, the transpalatal bar was installed to maintain the transverse stability. One year later, the plastic surgeon performed a total reconstruction of the lip and secondary palatoplasty, at this time a deepening of the vestibule funds was also performed. During the following three years, the patient missed follow-up.

At 9 years old, the patient returned to the odontology follow. In that moment, the patient had a mixed dentition, with the permanent central upper incisors erupted, bilateral agenesis of the lateral

upper incisors and the canines included with 1/3 of the root formed (Figure 2 – e), with the premaxilla projected forward and downward in relation to the posterior segments (Figure 2 – a, b, c), without seal lip (Figure 2 – d), but already demonstrating an improvement in the transverse distance of the maxilla due to maxillary expansion. The family related several social complaints, lack of motivation to go to school and difficult interaction with colleagues.



**Figure 1** – Follow-up of the 5-year-old patient: a) right extraoral profile photo; b) extraoral frontal profile photo; c) panoramic radiographic and d), e), f) intraoral photo.



**Figure 2** - Pre-operative monitoring, virtual planning and trans-operative photos of premaxillary repositioning surgery: a), b), c) intraoral photo; d) extraoral frontal profile photo; e) panoramic radiographic monitoring; f) computed tomography showing the anterior projection of the preoperative premaxilla; g) computed tomography showing the planning of anterior projection of the postoperative premaxilla; h) computed tomography showing the inferior projection of the preoperative premaxilla; i) computed tomography showing the planning of inferior projection of the postoperative premaxilla; j) and k) trans-operative photos showing the surgical repositioning of the pre-maxilla and internal rigid fixation with plates and screws.

It was decided to perform surgical repositioning the premaxilla using the Carlini & Biron technique<sup>8</sup>. To improve surgery prediction, to visualize premaxilla impaction and backward movement and to check bone interferences, virtual surgical planning was carried out using *nemotec software*. (Figure 2 – f, g, h, i). Then, a surgical guide was created, and an orthodontic arch was performed to stabilize maxilla after operation.

The surgery took place under general



anesthesia and nasotracheal intubation. After infiltration of lidocaine 2% and epinephrine 1:100.000 into the bottom of the maxillary vestibule, intraoral access was performed maintaining a vestibular pedicle in the premaxilla. On the buccal surface, access occurred bilaterally with relaxing incisions in the regions of teeth 55 and 65, followed by intrasulcular incisions and incisions in the cleft region. Afterwards, flap detachment and fistula closure were performed, separating the nasal cavity from the oral cavity bilaterally. Incisions were made in the palate to create the osteotomy lines, allowing the premaxilla to be repositioned.

The first osteotomy line was performed to separate the premaxilla from the vomer bone and the second, following virtual planning, was to remove almost 9mm of bone from the premaxilla vertically, this removed segment was particulate and later used for the autogenous graft in the fissure regions. The premaxilla was then repositioned using the surgical guide, being retreated antero-posteriorly by almost 5mm, and was fixed with 2 mini plates and 4 titanium screws (Figure 2 – j, k). The bone graft was positioned in the remaining gaps in the fissure region bilaterally, the flap was then repositioned, and the suture was performed with Monocryl 0.4 thread.

On the first postoperative day, the orthodontic arch was installed on the patient with the transpalatal bar to assist in stabilizing the premaxilla and occlusal leveling postoperatively. Eight days after surgery, the patient was in good general condition, with no spontaneous pain complaints and no signs or symptoms of infection. The mucous membranes were stained, and the premaxilla segment was stable with an orthodontic arch and sutures in position. On the 14th postoperative day, the edema had completely regressed, with no complaints, and it was already possible to see the improvement in facial appearance. The stitches were then removed.

During the six-month postoperative follow-up, the patient presented stable repositioning of the premaxilla, significant improvement in facial appearance, and improved chewing and speech function (figure 3 – a, c, d, e). The new tomographic examination demonstrated adequate positioning of the premaxilla and fixation material, however, low bone formation between the segments (figure 3 - b). Thus, a second surgery was performed to remove the fixation material and perform a new bone graft, now with iliac bone. The surgery was performed under general anesthesia, and graft removal was performed by an orthopedist. After intra-oral access in the bilateral cleft region, the fixation material was removed, and the autogenous bone was particularized and mixed with freeze-dried bone of bovine origin and positioned in the bone gaps.

During fifteen months post-operatively the patient presented a good anteroposterior positioning of the maxilla and lip seal (figure 4). Currently, the patient is post-operative two years from this second surgery and he is still undergoing orthodontic treatment with fixed appliances, is in the occlusal leveling phase and continues with the transpalatal bar (figure 4). Those responsible for the patient reported an improvement in the patient's quality of life, with improvements mainly in speech, chewing and the patient's social life.



**Figure 3** - Postoperative follow-up after premaxillary repositioning surgery: a) extraoral frontal profile photo; b) computed tomography monitoring and c), d), e) intraoral photo.



**Figure 4** – follow-up after bone graft surgery: a), d), e), f) intraoral photo; b) extraoral frontal profile photo and c) computed tomography monitoring.

## DISCUSSION

In this case, the repositioned premaxilla technique associated with secondary graft proved to be effective, without surgical or postoperative complications. With 15 months of postoperative follow-up, it is possible to notice functional and aesthetic improvements, improvements in speech, lip sealing and occlusion, leading to an increase in the patient's quality of life.

The surgical repositioning of the premaxilla must be done out with care, as complications may occur, such as: necrosis and loss of the premaxilla, infection and instability of the premaxilla associated with recurrence of fistulas<sup>10</sup>. In this case, virtual surgical planning was carried out, which increases the predictability of the surgery.

The literature describes two techniques to repositioning the premaxilla. The first one was described by Aburezq *et al.*, they preconize, at same time, the surgical repositioning of the premaxilla and the iliac bone grafting<sup>9</sup>. In four patients submitted of this procedure, no one has postoperative complications, and only one doesn't have the premaxilla consolidation<sup>9</sup>.

The second technique was described by Carlini *et al.*, they preconize, at same time, the surgical repositioning of the premaxilla and the iliac bone grafting with fixation using mini plates and screws<sup>4,8</sup>. In the 2009 article, they evaluated 50 patients and in 24 they used only mandibular symphysis bone grafting and in 26 used a mixed of iliac and mandibular symphysis bone grafting. Of these 50 patients, 48 cases (96%) the treatment achieved total graft integration and only 2 (4%) the treatment failed, due to necrosis of the premaxilla<sup>4</sup>. In the 2020 article, they evaluated 16 patients using only the iliac bone grafting and the treatment fail in only one patient<sup>8</sup>.

In this case report, we performed surgical repositioning of the premaxilla as described by Carlini *et al.*, but the bone graft using the iliac bone was performed at different surgical times because the alveolar process and the soft tissue that covered this region in our patient, it was not in ideal conditions, so we waited for the premaxilla to become stable.

Regarding the conditions of the reconstruction of alveolar process defects by secondary bone grafting, the ideal age in bilateral CLP patient varies between 8 and 12 years, before eruption of the permanent upper canines and when the canine root reaches 1/3 of its length, and while its crown is still covered with bone<sup>11,12</sup>. In this case report, this reconstruction was performed when the patient was 9 years old.

The secondary bone grafting could facility the post-orthodontic treatment and the post implants placement in the cleft region and improves a better osseus support on adjacent cleft structures like upper lip and nose<sup>13</sup>. To date, the autogenous graft is considered the gold standard for secondary grafts<sup>14</sup>.

Thus, this case report is relevant due to the description of the multidisciplinary approach of a challenging health condition, with a large time to following the patient, from five days of life until ten years old.

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## CONFLICT OF INTERESTS

The authors declare no conflict of interest.

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