



ISSN: 2230-9926

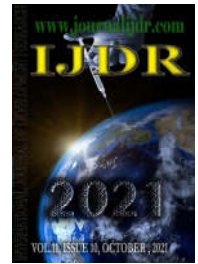
Available online at <http://www.journalijdr.com>

# IJDR

International Journal of Development Research

Vol. 11, Issue, 10, pp. 51099-51101, October, 2021

<https://doi.org/10.37118/ijdr.23019.10.2021>



RESEARCH ARTICLE

OPEN ACCESS

## MULTIDISCIPLINARY TREATMENT FOR UNILATERAL CLEFT PALATE REPAIR: 12-YEAR FOLLOW-UP

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### ARTICLE INFO

#### Article History:

Received 06<sup>th</sup> August, 2021  
Received in revised form  
17<sup>th</sup> September, 2021  
Accepted 08<sup>th</sup> October, 2021  
Published online 30<sup>th</sup> October, 2021

#### Key Words:

Bone Graft, Cleft Palate,  
Dental Implants, Dentistry.

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### ABSTRACT

Non syndromic cleft lip and palate is a common congenital anomaly. The treatment planning must be individualized for each patient, because there are individual needs that must be respected. The study reports a 12-year follow-up after the treatment of a young male patient with a right unilateral trans-foramen cleft, and absence maxillary left central incisor. The treatment firstly consisted of autogenous bone graft procedure to close the region, followed by insertion of an osseointegrated implant and rehabilitation with an implant supported metaloceramic dental prosthesis. The case presented satisfactory results after a time interval of over a decade after finishing the treatment.

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Citation: Idalisio Soares Aranha Neto, Carlos Henrique Bettoni Cruz e Castro, Amaro Ilidio Vespasiano Silva et al. "Multidisciplinary treatment for unilateral cleft palate repair: 12-year follow-up", *International Journal of Development Research*, 11, (10), 51099-51101.

## INTRODUCTION

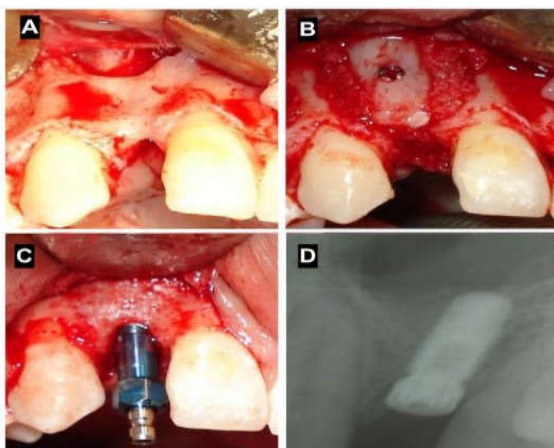
Non-syndromic cleft lip and palate is a multifactorial congenital deformity related to environmental and genetic factors (Lewis *et al.*, 2017). The condition is considered a public health concern, affecting on an average one in every 650 live births (Pucciarelli *et al.*, 2019). There are still large areas of the world that do not provide information or collect incomplete information about non-syndromic cleft lip and palate, which implies an underestimation of the overall prevalence (IPDTC, 2011). The classification of Spina introduced in 1972 is a simple method that uses the foramen incisor as the point of reference for classifying the types of cleft palate and/or lip. The classification denominates the groups as pre-foramen incisor, trans-foramen incisor and post-foramen incisor clefts, in addition to rare facial clefts. Group I of the pre-foramen incisor clefts are (incomplete) clefts of the lip, with or without the involvement of the alveolus (complete) and may be unilateral or bilateral.

Group II refers to trans-foramen clefts, affecting the lip, alveolar ridge and the entire palate, and the cleft may be unilateral or bilateral. Group III are post-foramen incisor clefts that involve only the region of the palate, and may be complete or incomplete. Group IV includes rare clefts of the face, involving oblique, transverse clefts of the bottom lip and nose, among other features (Spina *et al.*, 1972). The treatment of patients with clefts involves multidisciplinary work, initiated from the time of pre-natal diagnosis through to adult age. For each patient, the intervention must be individualized to enhance treatment outcomes (Lewis *et al.*, 2017). According to the recommendations of the American Society of Pediatrics, care of the patient begins right from the time of the first months of life, with cleft lip closure surgery (between 3 and 6 months of life). The cleft palate repair surgery is normally performed between 9 to 18 months of age. From 6 to 11 years of age, a visit to the orthodontist is indicated to evaluate the patient's orthodontic needs. Throughout the entire period of follow-up, periodical evaluations should be carried out for speech, sleep, psychological, nutritional, dental and auditory evaluations. In

addition, the patient's development and growth should be followed-up (Lewis *et al.*, 2017). After the age of 18 or the end of the bone growth phase, the second treatment stage begins, which consists of bone grafting procedure for alveolar reconstruction, soft tissue reconstruction and, subsequently, insertion of osseointegrated implants for functional and esthetic rehabilitation. The bone graft may be performed with autogenous bone taken from the iliac bone, tibia or mandibular symphysis. Biomaterials may also be used as bone substitutes (Bousdras *et al.*, 2015) and in cases requiring reconstructions of large areas with significant volumes of bone. The aim of the present study was to report the clinical case of a young patient who presented with unilateral trans-foramen cleft, in which the procedures of nasal communication closure were performed, rehabilitation with autogenous bone grafting and insertion of dental implant and followed-up for 12 years.

## CASE REPORT

The present case report was written following the Consensus-based Clinical Case Reporting Guideline Development (CARE) (Gagnier *et al.*, 2014). Informed consent was obtained from the patient. A male patient, Caucasian, 18 years old sought dental care complaining of absence of the left upper lateral incisor. The patient reported having undergone a bone graft procedure to close a cleft lip and palate during childhood without knowing how much time had occurred. After the initial anamnesis, the patient was referred to an assessment involving the maxillofacial surgery teams, implantodontists, plastic surgery, speech therapy and orthodontics for an integral treatment planning for the case. The extra oral exam evidenced only a small scar on the lip as a probable alteration generated by previous surgeries to correct nasolabial cleft, with a small loss of projection and motor skills in this area. Clinical intraoral examination evidenced a unilateral trans-foramen cleft leading to a communication between nasal and oral cavities. In addition, the absence of tooth 12 due to agenesis and a residual scar in the palate region and corresponding alveolar ridge were observed. The distal mesial space was preserved due to the use of a removable partial prosthesis. The patient also presented a small level of keratinized gingiva and bone loss in the vertical direction. There were no other occlusal changes, such as the absence of a vertical dimension, limited opening or occlusal trauma. Periapical and panoramic radiographs evidenced a low bone volume, which hindered the insertion of an osseointegrated implant. The treatment plan involved a bone graft procedure to close the oroantral communication and a subsequent installation of an osseointegrated implant in the site of tooth 12 for prosthetic rehabilitation with a single crown. The bone graft procedure was performed with autogenous bone removed from the iliac crest region by the orthopedics team under general anesthesia performed at a hospital operating room. Initially, extra oral asepsis was performed with iodized alcohol and intra oral asepsis with 0.12% chlorhexidinedigluconate. The autogenous (bicortical) graft was removed from the right iliac crest. The graft was particulate and inserted in the cleft palate (region of tooth 12), as well as a split flap for obliteration near the nasal cavity (Figure 1A-B).



**Figure 1.** Oral rehabilitation consisting of an autogenous bone graft to close oro-nasal communication and osseointegrated implant installation

After a four-month healing period, new radiographic images were taken and the maintenance of the bone graft was observed. Next, the osseointegrated implant installation was performed. After asepsis, anesthesia of the region was obtained with bilateral infraorbital and anterior superior alveolar nerve blocks, incisive foramen anesthesia, and infiltrative anesthesia in the periosteum. For these procedures, 2 tubes of Alphacaine 2% with epinephrine 1:100.000 (DFL®) were used. The incision was made in the crest of the alveolar ridge with detachment of the flap and visualization of the alveolar bone. The surgical drilling sequence followed a protocol recommended by the implant manufacturer (Straumann-Neodent®, Brazil). The cortical perforation respected the pre-established limits, preparing the middle distal space with orthodontic retaining with temporary teeth, which served as a surgical guide. This reverse planning allowed the proper three-dimensional positioning of the implants for the subsequent manufacture of the prosthetic crowns. After the initial drilling, the positioning and parallelism of the implants obtained through the surgical guide were checked. After the milling sequence was completed, 3.3 x 11.0 mm tapered implants were placed (Straumann-Neodent®, Brazil) (Figure 1C-D). After a healing period of 6 months, a metaloceramic crown was made and inserted over the implant (Figure 2A). Occlusal adjustments were made to obtain non-traumatic occlusal contacts (Figure 2B). After the completion of the surgical phase, the patient reported satisfaction with the treatment outcomes (Figure 2C). The patient was instructed to return periodically for reassessment, but he did not attend until returning after 12 years. The reassessment after 12 years confirmed the success of the treatment (Figure 3A-C) and the patient reported that he was still satisfied with the treatment outcomes.



**Figure 2.** Temporary prosthesis installation after osseointegrated implant procedure



**Figure 3.** Follow-up images after 12 year follow-up

## DISCUSSION

The case report in question presented the treatment of an 18-year-old Caucasian male patient with a right unilateral trans-foramen cleft. The patient had previously concluded the first stage of treatment with the closure of the cleft in childhood, as recommended (Lewis *et al.*, 2017; Jabbari *et al.*, 2016). The patient sought dental care with the chief complaint being the absence of the maxillary left lateral incisor. A retrospective analysis of 207 panoramic radiographs of Brazilian patients with cleft lip and/or palate not associated with syndromes showed that 75.4% of patients with clefts presented some type of dental anomaly, and that among them, the absence of teeth was the most prevalent (Sá *et al.*, 2016). This information was in agreement with the personal history of the patient described herein. The lack of a tooth, especially in the anterior region, can lead to speech and chewing disorders, and high-level aesthetic problems, thus having a major negative impact on the oral health-related quality of life (OHRQoL) (Silva-Rodrigues *et al.*, 2018). In addition, patients with dental agenesis may present changes in the sagittal growth of the

maxilla in relation to the positioning of the lower incisors (Herrera-Atoche *et al.*, 2020), what makes this condition indicative of multidisciplinary treatment as soon as possible. The second stage of rehabilitation in patients with cleft lip and or palate consists of alveolar bone grafting and replacement of the missing tooth to re-establish the patient's esthetics and function. The ideal time for performing the graft is yet to be a consensus, and may be a primary graft together with surgery for closure of the cleft; or secondary, after initial closure surgery (Bousdras *et al.*, 2015). In the clinical and radiographic exams of the patient in question, a reduced bone volume was found in the region of the missing tooth, so a new bone graft procedure was indicated. Bousdras *et al.* (2015) suggested that grafts made with bone from the iliac crest were the gold standard for alveolar bone reconstruction due a large available bone to be removed with a relatively easy access, and the presence osteogenic cells that would have a positive influence on the period of healing after the surgical procedure.

In the present case, due to the large bone defect, a bone graft was removed from the right iliac crest. Removal in intraoral areas was not considered due to the limited availability of bone. At present, biomaterials are increasingly being used as a successful alternative to autogenous bone grafts (Shamsoddin *et al.*, 2019). In addition, a systematic review (Wu *et al.*, 2018) showed that bone morphogenetic proteins associated with a collagen sponge had a satisfactory effect for alveolar grafting compared with autogenous material obtained from the iliac crest. It is valid to point out that the surgical phase in this present case was performed 12 years ago, and at the time of treatment planning, these options were not yet very accessible, particularly in public health services. Some authors argue that the implant insertion should be performed in up to 6 months after the graft procedure (Bousdras *et al.*, 2015). Although it is also possible to perform an immediate insertion, there are associated risks such as the occurrence of height bone graft loss. In the present case, the clinical decision-making was to insert the dental implant four months after performing the bone graft, which is in agreement with the recommendations in the literature (Misch, 2011). Also, an immediate insertion of a temporary acrylic implant-supported dental prosthesis was carried out. After 6 months of osseointegration, a definitive metaloceramic dental prosthesis was made. The multidisciplinary rehabilitation achieved with the closure the cleft, bone volume gain with autogenous bone graft procedure, placement of an osseointegrated dental implant, and fabrication of an implant-supported dental prosthesis promoted an improvement in both function and esthetics of the patient. As a result, the patient reported a great satisfaction with the treatment outcomes. In this context, the insertion of implants is considered the gold standard in the process of esthetic rehabilitation of patients with clefts, thus maximizing the patient's satisfaction with the treatment (Papi *et al.*, 2015). Moreover, it is reported that restoring the patient's OHRQoL by means of rehabilitation was a challenging, but a possible and desirable task. Few studies have been able to achieve such a long follow-up after conclusion of a case. Twelve years after the treatment, the patient still presents a good bone level in the region where the bone graft was performed, and the requisites of esthetics and function are in satisfactory conditions. The patient also demonstrates a high satisfaction with treatment outcomes. Other studies, such as that of Jabbari *et al.* (2016) presented follow-up of patients right from the time of the first interventions in childhood, with closure of the cleft, through to the beginning of the second stage of treatment. In spite of presenting a 10-year period of follow-up, the focus differed from that of the case reported herein, in which the main aim was to emphasize the stability of the prosthetic and surgical treatment more than one decade after their conclusion.

## CONCLUSION

In cases of cleft lip and palate, a multidisciplinary approach involving maxilla ofacial and plastic surgeons, implantodontists, speech the rapist and orthodontists is crucial for the planning and execution of the treatment.

**Conflict of Interest:** The authors declare that there is no conflict of interest regarding the publication of this article.

**Acknowledgments:** The authors thank the "Pontifícia Universidade Católica de Minas Gerais" – PUC Minas (Pontifical Catholic University of Minas Gerais); "Coordenação de Aperfeiçoamento de Pessoal de Nível Superior" – CAPES (Coordination for the Improvement of Higher Education Personnel); and the "Fundação de Amparo à Pesquisa do Estado de Minas Gerais" – FAPEMIG (Minas Gerais State Research Support Foundation) for supporting the research.

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