

Aesthetic satisfaction in lip and palate clefts: a comparative study between secondary and tertiary bone grafting

E. Brauner¹, F. De Angelis¹, S. Jamshir¹, S. Mezi³, R. C. Tiroli², G. Pompa¹, A. Quarato², S. Di Carlo¹

¹Department of Oral And Maxillo-Facial Sciences, Sapienza University of Rome; ²Private Practice; ³Dipartimento di Scienze radiologiche oncologiche ed anatomopatologiche, Rome

Abstract

Lip and palate cleft represent one of the most frequently occurring congenital deformity, which includes dental anomalies, such as variation in tooth number and position. In case of hypodontia implant-prosthetic rehabilitation offers significant advantages in terms of function, aesthetics and quality of life and bone graft is usually needed. Secondary bone grafting, generally performed in the mixed dentition phase (years 8-11) seems to be the most successful method to allow for rehabilitation. It's often necessary to perform a tertiary bone grafting in adult age in order to achieve better bone quantity and quality before implant placement. Aim of this retrospective study was to evaluate the aesthetic perception that patients had of themselves comparing dental implants placed in tertiary grafted alveolar cleft sites with a previous secondary grafting to only secondary grafting. Between 2009 and 2012, fourteen alveolar cleft were treated with implant rehabilitation and eleven of them received tertiary bone grafting six months prior to implant placement. All patients were questioned to give a score from 1 to 10 their aesthetic satisfaction of their smile before and after implant rehabilitation and during pre-surgery provisional rehabilitation. At the end of their prosthesis rehabilitation patients who received tertiary bone grafting resulted more satisfied than those who had secondary bone grafting only (9.5 vs 8). *Clin Ter 2018; 169(2):e62-66. doi: 10.7417/CT.2018.2055*

Key words: Lip Cleft, Palate Cleft, Bone Graft

Introduction

Cleft lip and palate (CLP) is among the most frequent congenital malformations, its incidence varying from 1 to 7/1000 newborns (1). Failure of fusion of the maxillary and medial nasal prominences or between the palatal processes results in clefts of varying extent, unilaterally or bilaterally. Compared to general population, CLP is associated with a higher percentage of dental anomalies, such as variation in tooth number and position, well as feeding and speaking clearly (2). There is a big variety of CLP with a big range of severity. We can classify them by means of different indices created for CLP cases: the GOSLON Yardstick is the most prevalent one, possibly due to a longer time in use, while

the MHB outperformed the rest of the indices for an ideal index (1). It is the authors' recommendations that the MHB Index be used to assess the malocclusions of all clefts of the lip and/or palate of all ages and to standardize the measurement of outcomes in cleft lip and palate patients' malocclusions in order to facilitate international inter-centre studies and allow for the optimization of cleft treatment protocols (1).

The prevalence of hypodontia in cleft lip and palate seems to increase with the cleft severity (2). Shapira found that hypodontia was present in 77% of non-syndromic cleft children (3). In general, tooth missing is about 10 times more frequent on the cleft side, with left prominence (4, 5).

The aim of current treatment protocols is the restoration of bone and soft tissues continuity and oral rehabilitation, in order to return to a correct speech and masticatory function, as well as an aesthetically satisfactory result. Cleft patients are usually treated by a combined approach, which involves the collaboration of maxillofacial surgeons, orthodontists, speech therapists, oral surgeons and prosthodontists (6, 7). Rehabilitation should be planned, when possible, before surgical treatment, in order to cooperate with the maxillofacial surgeon in choosing the most appropriate restorative treatment (8).

Primary grafting includes palate closure (after lip closure) by the age of 1 year, but today it isn't the golden choice because it leads to unfavorable growth compared to later conventional graftings (9).

Secondary bone grafting, suggested by Boyne and Sands, is usually performed during the mixed dentition (years 8-11) and it seems to be a successful method to allow for rehabilitation of alveolar cleft patients (10,11). It ensures functional and aesthetic results, re-establishing the alveolar bone profile and allowing the closure of oro-nasal communications.

Combined to orthodontic treatment, the grafting should eventually contribute to obtain a uniform upper arch with adequate sagittal and transversal width (12), the stabilization of the jaw segments, and osseous support for the alar base (10). With the result of a stronger alveolar bone in the area of the teeth neighboring the cleft, the bone graft should also permit movement of adjacent teeth, and the eruption of them through the transplant into the cleft (12). According to literature, the best results of alveolar bone grafting in cases

of secondary osteoplasty were obtained when the lateral incisor or canine had grown into the transplant and had led to a functional stress of the transplanted bone (10). These preconditions allow normal possible dentition to develop without any skeletal disharmonies and to form a regular anterior tooth overjet and overbite (13).

When one or more teeth are missing in the cleft area, implant placement in adulthood is the most viable option in terms of function and aesthetics, preferable to a removable prosthesis, fixed on natural teeth prosthesis or adhesive prosthesis (14): implants rehabilitation offers aesthetical advantages, fits more comfortably young patients, preserves healthy tissue of the adjacent teeth, functionally loads the bone graft, thus preventing its resorption (15).

As the secondary bone grafting is usually performed between 8 and 11 years old, and in any case before canine eruption, whereas implants should not be placed in growing phase, several years elapse before implant rehabilitation. In agreement with Steinberg et al. (1999), it is therefore often necessary to perform a new graft (tertiary bone grafting) before implant placement, as to obtain an adequate space for correct implant placement and an aesthetically satisfactory contour of the alveolar process (16).

Reconstructed patients involve multidisciplinary approach and are more difficult to rehabilitate; treatment is also influenced by other clinical parameters: gingival tissue could need a periodontal surgery intervention and soft tissue damage could require a plastic surgery (17,18).

There are many literature investigations about secondary and tertiary bone grafting in CLP cases

According to the article by Takahash et al., authors reported their experience with 16 patients (8 males and 8 females) who were submitted to secondary grafting of alveoli: they placed dental implant in grafted alveoli using particulate cancellous bone and marrow from the iliac crest (19). Mean age at first graft surgery was 14.1 ± 5.3 years, while mean age at first implant placement was 19.1 ± 4.7 years. Authors wanted to demonstrate that dental implants are beneficial for maintenance of the grafted bone. They managed this purpose by making radiological evaluations of the marginal

bone and the interdental alveolar bone height in a follow-up period of an average of 8.6 ± 0.6 years.

As regard investigations about tertiary bone grating, most of them are made in adult patients who received this treatment as second surgery after the starting lip closure: they directly had tertiary grafting without secondary one.

This is the case, for exemple, of the patients studied in the article by Ronchi, Chiapasco et al. They led the surgical and prosthetic care of three adult patients affected by sequelae of cleft lip and palate, with residual alveolar cleft and absence of maxillary anterior teeth. In their experience, patients were treated with tertiary bone grafting (as second surgery after palate closure) of the alveolar cleft followed by the insertion of endosseous titanium plasma-sprayed implants after 6 or 12 months. According to these authors, dental prostheses supported by endosseous implants in grafted alveolar clefts are a very reliable possibility in dental rehabilitation of this malformation (20).

Further evidences about tertiary graftings benefits were documented in a retrospective study by Dempf et Al. In this case 91 patients (2 with unilateral and 49 with bilateral clefts) received a total of 140 osteoplasties and 47 of them were tertiary osteoplasties planned in order to place an endosseous implant for prosthodontic indications. Approximately good results were to be found in tertiary osteoplasty when the transplanted bone had been stressed functionally through a dental implant (10).

The aim of this retrospective study was to evaluate the outcome of dental implants placed in tertiary grafted alveolar cleft sites with a previous secondary grafting compared to only secondary grafting, particularly focusing on the aesthetic perception that patients had of themselves before and after the treatment.

Methods

Between 2009 and 2012, fourteen alveolar cleft patients (nine female, five male, aged from 18 to 22, mean age 19) were treated for tooth missing in the cleft area with implant rehabilitation, for a total of 16 implants placed (table 1). All patients involved in this study had received secondary

Table 1. Patient and treatment characteristics

N°	Sex	Age (years)	Implant location	Tertiary grafting	Implant brand	Aesthetic satisfactory (range 1-10) at the end
1	F	20	22, 23	YES	TM Zimmer	9
2	F	20	22	NO		7
3	M	20	23	YES		9
4	F	22	13	YES		10
5	F	20	21	YES		9
6	M	18	22, 23	YES		10
7	F	19	12	YES		10
8	M	21	13	YES		9
9	M	19	21	YES		9
10	F	18	12	NO		8
11	F	20	22	YES		10
12	M	19	22	NO		9
13	F	20	22	NO		8
14	F	21	23	YES	TM Zimmer	10

bone grafting between 8 and 11 years old. This study was carried out in accordance with the ethical standards of the 1964 Declaration of Helsinki and informed written consent was obtained from each patient.

All of the patients had indication to receive a further grafting in order to improve their bone quantity and quality, but not all of them wanted to be operated again: of the fourteen ones selected, eleven received tertiary bone grafting six months prior to implant placement.

The tertiary bone graftings had been realised with heterologous cancellous particulate bone and collagen resorbable membranes and a tension free suture was always performed over the graft. The site was accessed again after a healing period of six months for implant placement, in order to allow proper bone maturation.

One hour before surgery (bone graft and implant placement), prophylactic antibiotic therapy was administered to the patients: 2g of amoxicillin and clavulanic acid or, in case of penicillin allergy, 500 mg of azithromycin. Chlorhexidine digluconate 0,12% mouthwash was prescribed every day for seven days after surgery. Patients were provided with written instructions for oral hygiene and were advised to follow a soft diet for four to five days postoperative.

All implants were placed after flap elevation and were then submerged and allowed to heal from four to six months. At surgical uncovering, healing collars were placed and left in place for three weeks. Temporary crowns were then cemented and left in place for one month to allow soft tissue maturation. The restorations were finalized with ceramometal-cemented crowns.

Implant rehabilitation evaluation was performed with clinical and radiographical monitoring during recall visits.

In addition, all patients were questioned to give a score from 1 to 10 (1 for totally unsatisfactory, 10 for totally satisfactory) to their aesthetic satisfaction of their smile before and after implant rehabilitation and during pre-surgery provisional rehabilitation (partial removable prosthesis or adhesive type Maryland Bridge) (Tab. 2). The null hypotheses of independence of the results were tested by Student t test.

Results

Post-operative course was uneventful in all cases. No implant was lost before loading or presented clinical or radiological signs of failure during recall visits (minimum follow-up was of two years). It's important to considerate some factors which had influenced the success of the graftings (21): positive predictive factors are, for exemple, a generous transplant volume, 6 months latency, a sufficient implant dimension, an early adulthood; negative predictive factors are spongiosa or milled-bone transplants, dehiscence, smoking, and anorexia.

In patients who did not undergo tertiary bone grafting implant positioning was strongly influenced by bone quantity (22).

The average score given by the fourteen patients to their smile before any prosthetic rehabilitation was 4.6 (M=5.8, F=4). After pre-surgery rehabilitation (partial removable prosthesis or adhesive type Maryland Bridge), average score

was 6.7 (M=7.2, F=6.4), and after implant rehabilitation was 9 (M=9.2, F=9). The results were statistically significant.

Although average score after implant rehabilitation was 9, it was calculated a statistically significant difference between patients who underwent tertiary grafting (score 9.5) and those who did not (score 8) ($p<0,01$)

Discussion

According to systematic reviews, success rate for implants placed in native bone is of 97% after 7 years (23, 24). Reported success rate for implants placed in the area of an alveolar cleft after bone grafting is from 80% to 90% (25-26): when one or more teeth are missing in the cleft area, implant placement in adulthood is the better option for function and aesthetics, contrasting bone grafting areas resorption (15, 27). Good stability of the denture is provided and speech, mastication and deglutition are improved²⁸. In conformity with the results of this study, we can affirm that at the end of their prosthesis rehabilitation patients who received tertiary bone grafting are more satisfied than those who had secondary bone grafting only (9.5 vs 8): we can link tertiary bone grafting with more final bone quantity and, consequently, more aesthetics. 4 patients of the total 10 with tertiary grafting expressed a maximum level of aesthetical satisfaction, as a 10/10, while only one from second grafting patients was satisfied as 9, being lower the others values. Then, we found the same level of satisfaction for both the women and the men group at the end of their prosthetic rehabilitation over implants.

The tertiary grafting procedure increased the vertical alveolar height, in order to obtain an adequate positioning of implants which can assure more quality in the prosthesis and a correct smile line, lower and more aesthetic than before (12). This effect is fundamental especially in the cases in which the vertical growth deficit was done to the missing lateral incisor (Fig. 1,2): in the group of tertiary

Table 2. Patient aesthetic satisfactory, scored from 1 to 10 (1., 10)

N°	Before any dental rehabilitation	Rehabilitation before implants	Implant supported rehabilitation
1	3	5	9
2	4	6	7
3	6	7	9
4	4	6	10
5	3	5	9
6	5	6	10
7	5	8	10
8	6	8	9
9	5	7	9
10	5	7	8
11	5	8	10
12	5	8	9
13	5	6	8
14	4	7	10



Fig. 1



Fig. 2

osteoplasties, we found less cases of incisal infraocclusion. In our opinion, that is due to the fact that less height of the bone can be compensated easier by prosthetic mean9. Also the gingival contour became more regular and natural because of hard and soft tissues disponibility, even if it takes time until the final contours are reached because of the scarring of the mucosal tissues resulting from the previous operations. On the contrary, patients with only secondary grafting presented more irregularities in the gingival line and interdental papillae are frequently absent, but a low lip line luckily masked this characteristic (12, 24). Then, we have to consider that in LPC patients, aesthetic impact is mainly localised around the nasal aperture on the cleft side, resulting in displaced nasal cartilages, and hypoplasia of the pyriform rim, that affects the nasolabial complex and results in significant nasal asymmetry (9). Facial deformity and the most important defects of the nasolabial complex can be corrected with rhynoplasties ad oral secondary bone graftings, but they cannot be solved completely: the canina

and lateral incisor area, after secondary graftings, maintain some depression and the tertiary grafting can provide a solution by filling it with bone (29) (Fig.1,2). Consequently, patients are likely to obtain a more natural profile of the bone prominence in the cleft area.

In accordance with our retrospective study, we can affirm that the patients who received tertiary grafting have a stronger predictability of the aesthetics satisfaction. In this study we don't provide objective information about the quantity and quality differences of regenerated bone between the two groups, but we can say that tertiary grafting is the most predictable outcome as aesthetics. Economic cost of the surgery, rejection of the patient to be re-operated and longer rehabilitation time represent some of the disadvantages of this procedure (10, 30-33).

These are the reasons why 3 of the 14 patients rejected the extra surgery.

An important limit of this retrospective study is that patients' follow up is very short, minimum 2 years but maximum 5 years, with high variability for each case. Bone reabsorption after grafting is something important to be observed in the long term: the aesthetics of the prosthesis can be influenced and patients' satisfaction could change in a longer follow up. Furthermore, donor site morbidity, local growth, and tooth breakthrough require additional observation in prospective studies when implant insertion should directly follow the growth spurt (21).

Conclusion

The small number of this study limits firm conclusions, and further investigation will be needed. Anyway, the results suggest that although secondary grafting is sufficient in a selected number of cases, the best implant positioning and soft tissue adaptation is achieved with additional tertiary bone grafting, thus allowing better aesthetic outcomes and patients' satisfaction. We are going to follow up these patients and to analyse further outcomes in the long term.

References

1. Altalibi M, Saltaji H, Edwards R, Major PW, Flores-Mir C. Indices to assess malocclusions in patients with cleft lip and palate. *Eur J Orthod* 2013; 35:772-82
2. Ranta R. A review of tooth formation in children with cleft lip/palate. *Am J Orthod Dentofacial Orthop* 1986; 90:11-8
3. Shapira Y, Lubit E, Kuflinec MM. Hypodontia in children with various types of clefts. *Angle Orthod* 2000; 70(1):16-21
4. Shapira Y, Lubit E, Kuflinec MM. Congenitally missing second premolars in cleft lip and cleft palate children. *Am J Orthod Dentofacial Orthop* 1999; 115:396-400
5. Oberoi S, Vargervik K. Hypoplasia and hypodontia in Van der Woude syndrome. *Cleft Palate Craniofac* 2005; 42:459-66
6. Pena WA, Vargervik K, Sharma A, et al. The role of endosseous implants in the management of alveolar clefts. *Pediatr Dent* 2009; 31(4):329-33
7. Brauner E, Jamshir S, Papi P, et al. Obturator prostheses in post-oncological maxillofacial patients: our experience

- Senses Sci 2014; 1 (4):119-22
8. Brauner E, Valentini, S. Jamshir, A. Battisti, G. Guarino, A. Cassoni, G. Gaimari, M.T. Fadda, S. Di Carlo, G. Pompa. Two clinical cases of prosthetic rehabilitation after a tumor of the upper maxilla. *Eur Rev Med Pharmacol Sci* 2012.16(13): 1882-1890.
 9. Gillgrass TJ, MacDonald JP, Mossey PA, et al. Impact of Secondary Alveolar Bone Grafting on Cleft Lip and Palate: A literature review, *South Eur JOrthod Dentofacial Res* 2014;1:15-22.
 10. Dempf R, Teltzrow T, Kramer FJ, et al. Alveolar bone grafting in patients with complete clefts: A comparative study between secondary and tertiary bone grafting. *Cleft Palate Craniofac J* 2002; 39:18-25
 11. Duskova M, Kotova M, Sedlackova K, et al. Bone reconstruction of the maxillary alveolus for subsequent insertion of a dental implant in patients with cleft lip and palate. *J Craniofac Surg* 2007; 18:630-8
 12. Cune MS, Meijer GJ, Koole R. Anterior tooth replacement with implants in grafted alveolar cleft sites: a case series. *Clin. Oral Impl Res* 2004; 15:616-24
 13. Cassoni A, Valentini V, Della Monaca M, et al. keratocystic odontogenic tumor surgical management: retrospective analysis on 77 patients. *European Journal Of Inflammation* 2014; 12: 1,209-215
 14. Brauner E, Jamshir S, G. Guarino, et al. Pleomorphic adenoma rehabilitative treatment in growing up patient: a 20-years follow-up. *European review for medical and pharmacological sciences*. 2016; 20: 3523-7
 15. Honma K, Kobayashi T, Nakajima T, et al. Computed tomographic evaluation of bone formation after secondary bone grafting of alveolar clefts. *Journal of Oral and Maxillofacial Surgery* 1999; 57:1209-13
 16. Steinberg B, Padwa B, Boyne P, et al. State of the Art in Oral and Maxillofacial Surgery: Treatment of Maxillary Hypoplasia and Anterior Palatal and Alveolar Clefts, *The Cleft Palate-Craniofacial Journal* 1999;36(4):283-291. V
 17. Brauner E, Pompa G, Quarato A, et al. Maxillofacial prosthesis in dentofacial traumas: A retrospective clinical study and introduction of new classification method. *BioMed Research International* 2017; Article ID 8136878,1-8
 18. Valentini V, Terenzi V, Cassoni A, et al. Giant cell lesion or Langerhans' cell histiocytosis of the mandible? A case report. *European J of inflammation*. 10(1):159-164
 19. Takahashi T, Inai T, Kochi S, Fukuda M, Yamaguchi T, Matsui K, Echigo S, Watanabe M. Long-term follow-up of dental implants placed in a grafted alveolar cleft: evaluation of alveolar bone height. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2008; 105(3):297-302
 20. Ronchi P, Chiapasco M, Frattini D. Endosseous implants for prosthetic rehabilitation in bone grafted alveolar clefts. *J Cranio Maxillofac Surg* 1995; 23:382-6
 21. Constantin A. Landes, Implant-Borne Prosthetic Rehabilitation of Bone-Grafted Cleft Versus Traumatic Anterior Maxillary Defects *J of Oral and Maxillofac Surg* 2006; 64(2):297-307
 22. Brauner E, Cassoni, A Battisti, et al. Prosthetic rehabilitation in post-oncological patients: report of two cases. *Annali di Stomatologia* 2010; I (1):19-25
 23. Creugers NH1, Kreulen CM, Snoek PA, et al. A systematic review of single-tooth restorations supported by implants. *J Dent* 2000; 28(4):209-17
 24. Krennmair G1, Schmidinger S, Waldenberger O. Single-tooth replacement with the Frialit-2 system: a retrospective clinical analysis of 146 implants. *Int J Oral Maxillofac Implants*. 2002 Jan-Feb;17(1):78-85
 25. Carmichael RP, Sándor GK. Use of dental implants in the management of cleft lip and palate. *Atlas Oral Maxillofac Surg Clin North Am* 2008; 16(1):61-82
 26. Takahashi T, Fukuda M, Yamaguchi T, Kochi S, Inai T, Watanabe M, Echigo S. Use of an osseointegrated implant for dental rehabilitation after cleft repair by periosteoplasty: a case report. *Cleft Palate Craniofac J*. 1997 May;34(3):268-71.
 27. Pompa G, Brauner E, Jamshir S, et al. Quality of life in patients rehabilitated with palatal obturator without reconstruction versus fixed implant-prosthesis after reconstruction of maxillectomy defects. *J Int Dent Med Res* 2017; 10: (1): 1-8
 28. Brauner E, Valentini V, Guarino G, et al. Osteoradionecrosis Of A Mandible: A case report of implant-supported rehabilitation. *European Journal Of Inflammation* 2013; Vol. Ii: 2: 565-71
 29. Pagnoni M, Amodeo G, Fadda, et al. Juvenile Idiopathic/Rheumatoid Arthritis and Orthognatic Surgery Without Mandibular Osteotomies in the Remittent Phase. *Craniofacial Surgery J*. 2013; 24(6):1940-5
 30. De Felice F, de Vincentiis M, Valentini V, et al. Management of salivary gland malignant tumor: the Policlinico Umberto I, "Sapienza" University of Rome Head and Neck Unit clinical recommendations. *Crit Rev Oncol Hematol* 2017; 120:93-7
 31. Mezi S, Chiappetta C, Carletti R, et al. Clinical significance of epithelial-to-mesenchymal transition in laryngeal carcinoma: Its role in the different subsites. *Head Neck*. 2017 Sep;39(9):1806-1818. doi: 10.1002/hed.24838. Epub 2017 May 31.
 32. De Felice F, de Vincentiis M, Valentini V, et al. Follow-up program in head and neck cancer. *Crit Rev Oncol Hematol*. 2017 May;113:151-155. doi: 10.1016/j.critrevonc.2017.03.012. Epub 2017 Mar 14. Review
 33. Brauner E, Quarato A, De Angels F, et al. Prosthetic rehabilitation involving the use of implants following a fibula free flap reconstruction in the treatment of Osteosarcoma of the maxilla: A case report. *Clin. Ter*. 2017; 168:6: e392-63