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Ceramic implant-supported fixed prostheses in a patient with ectodermal dysplasia and cleft lip and palate: a case report

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ABSTRACT

Background Both cleft lip and palate (CLP) and ectodermal dysplasia (ED) are congenital conditions that hamper conventional prosthetic rehabilitation. CLP adult patients usually have sequelae and atrophy of the maxilla. In addition, dental malformation and positioning due to ED often limit the prosthetic alternatives for oral rehabilitation.

Case report This clinical report describes a challenging case of a patient with combination of CLP and ED, who was treated with dental implants and ceramic fixed prostheses, according to the patient's desire to have a pleasant smile and the comfort of fixed dentures.

Conclusion This report shows that ceramic implant-supported fixed prostheses are predictably safe when sufficient alveolar bone is present in a patient with both CLP and ED, thus restoring aesthetics and function.

KEYWORDS Complete denture, Dental esthetics, Fixed denture, Congenital abnormalities, Quality of life.

INTRODUCTION

Cleft lip and palate (CLP) is one of the most frequent congenital defects involving face and maxilla (1,2,3), with a prevalence rate of approximately 0.73 to 2.14 in every thousand births and significant variation due to ethnicity and location (1,3,4). The rehabilitation of CLP patients depends on the type and extension of craniofacial anomaly and requires an interdisciplinary approach since the first days of life till adulthood (5-7). Ectodermal dysplasia (ED) is a group of syndromes in which two or more anatomic structures derived from the ectoderm present development disorders. ED affects approximately 7 in every 10,000 births (8). The patients with ED may have cleft lip and/or palate in addition to other developmental disorders in structures originating from the ectoderm (teeth, hair, nails, body hair) (9).

The combination of CLP and ED poses a great challenge for prosthetic rehabilitation because of patient's atrophic maxilla and developmental abnormalities, such as dental malformation (conoid teeth), hypodontia or anodontia, which impairs occlusion and masticatory function (10-13). Bone usually grows normally, except for the alveolar process due to the absence of multiple teeth, which leads to a reduced vertical dimension of occlusion (VDO) (9, 14). The use of dental implants and prosthetic modifications circumvent the limitations of conventional prostheses in patients with reduced VDO, malformation, and missing teeth (7, 14, 15).

This article reports and discusses the oral rehabilitation with ceramic implant-supported fixed prostheses in a patient with CLP and ED.

CASE REPORT

A 21-year-old male patient with CLP and ED sought treatment at the Outpatient Prosthodontics Clinic of the Pontifical Catholic University of Rio Grande do Sul, in Porto Alegre, Brazil. His initial oral condition was categorized as Class IV, according to the ACP Classification System for partial edentulism. The patient had undergone orthognathic surgery before starting the oral rehabilitation treatment at the Dental School and was willing to improve his oral condition showing missing teeth and dental malformation.

The orthognathic surgery consisted of maxillary advancement by distraction technique and mandibular retrusion. After Le Fort I maxillary osteotomy, the

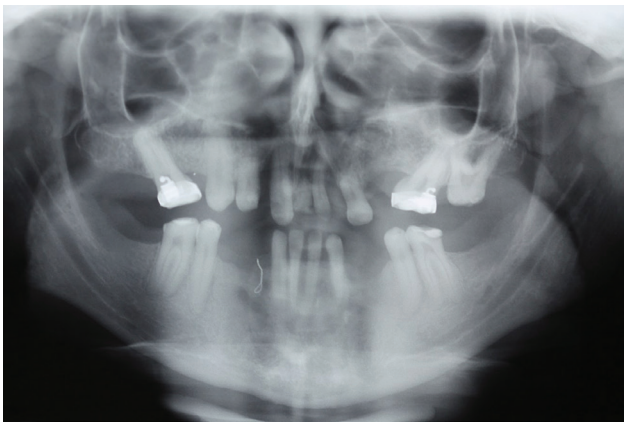
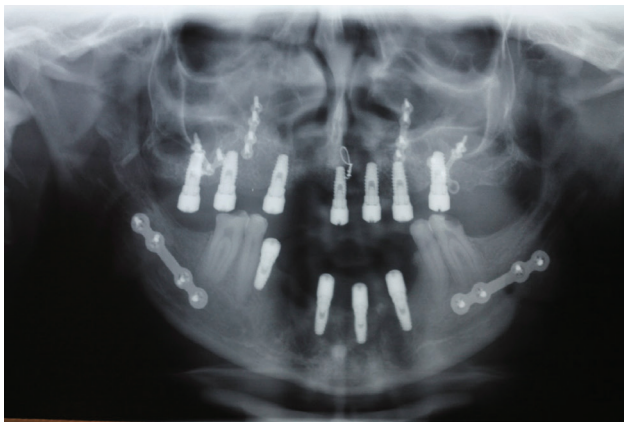


FIG. 1 Initial panoramic radiograph before teeth extraction.

advanced maxilla was fixed with miniplates in the new position, and a mandibular setback surgery was performed to obtain a normal occlusal relation, simultaneously (Fig. 1). Due to the bad conditions and position of the remaining teeth, some could not support a fixed prosthesis. In the mandible, the second premolars and first molars were maintained. Since the patient had a good amount of alveolar bone, the prosthetic planning was a maxillary full-arch denture and a mandibular partial-arch denture, both supported by dental implants. In a hospital operating room and under general anesthesia, multiple extractions of all maxillary teeth and mandibular incisors were performed. Immediately, seven implants were placed in the maxilla and four implants in the mandible (Osseotite Tappered Implant 4x10mm Biomet 3I, Warsaw, IN, USA), and the healing abutments were installed (Fig. 2). The patient used temporary removable prostheses with a soft liner (Silagum Confort Relining, DMG, Hamburg, Germany) during the osseointegration period in order not to overload the implants. One implant, placed at the region of the maxillary left central incisor, which is an area of the pre-maxilla with mobility due to CLP and needed previous bone graft, was lost after five months. Nevertheless, the original prosthetic plan was followed with six implants in the maxilla.

Six months after implant surgery, temporary acrylic implant-fixed prostheses with metal reinforcement were fabricated. The abutments were installed with a torque of 20 Ncm, and an open tray impression technique was used with impression copings union to maximize fit and passivity. A wax try in was performed (Fig. 3), and the vertical dimension of rest (VDR) was measured by using a Willis compass; VDR was subtracted 3 mm to achieve a standard VDO. Additionally, phonetic and aesthetic assessments were performed to check the established VDO. The wax upper-anterior volume was adjusted to the patient's hypotonic lip and atrophic maxilla. An occlusal record was obtained to mount the work casts in a semi-adjustable articulator and to assemble the artificial



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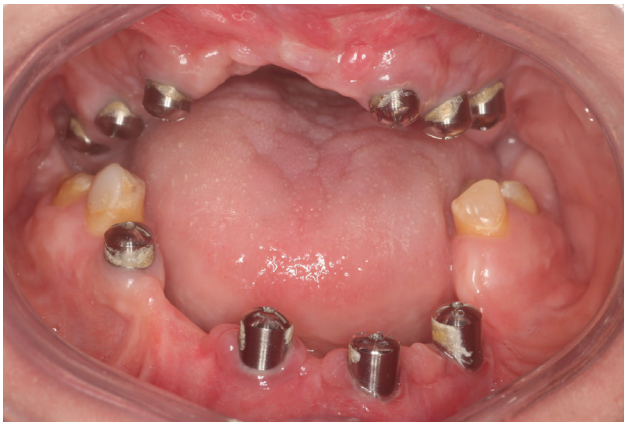

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FIG. 2 A Panoramic radiograph after installation of implants in maxilla and mandible.
B. Patient with implants and healing abutments installed, before prosthetic rehabilitation.

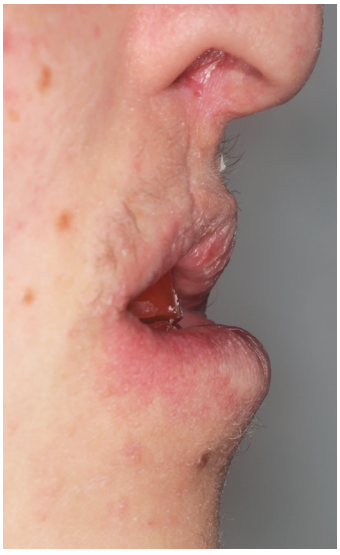
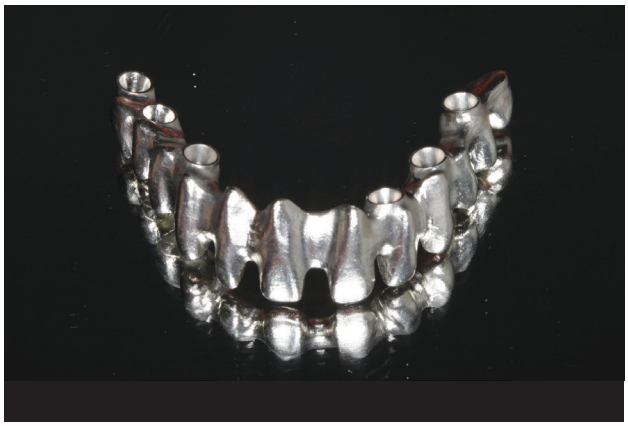


FIG. 3 Wax try-in to establish vertical dimension of occlusion and lip support (note that even after orthognathic surgery for correction of the maxillary atrophy, the patient still had a lip deficiency).

teeth in laboratory. After clinical testing for occlusion, aesthetics, and patient's acceptance, the temporary fixed prostheses were finished and installed in mouth with a torque of 10 Ncm on the prosthetic screws. The patient was followed for 11 months to allow adaptation to the



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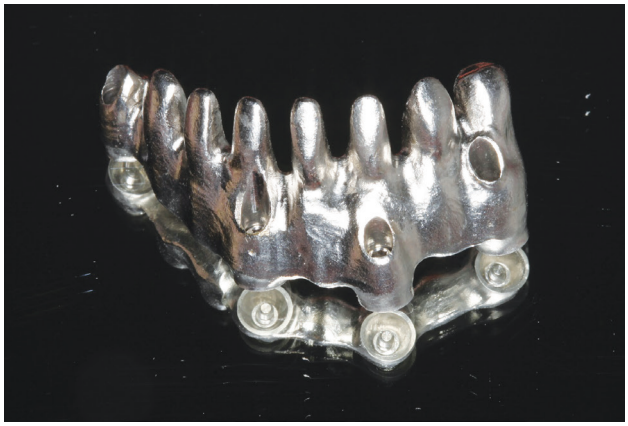

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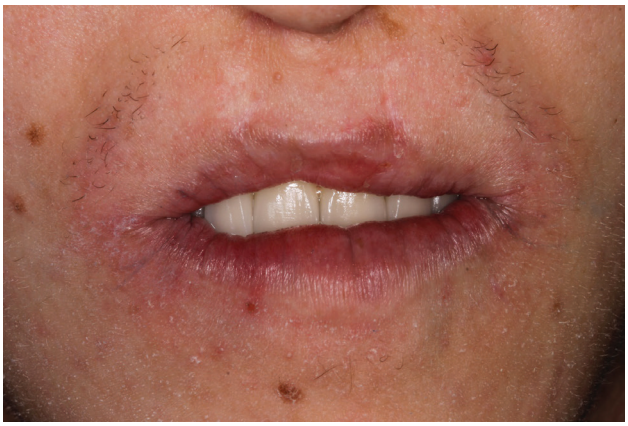
FIG. 4 Metal framework before ceramic coating. A. Maxillary full-arch implant-supported fixed denture. B. Mandibular partial-arch implant-supported fixed denture.



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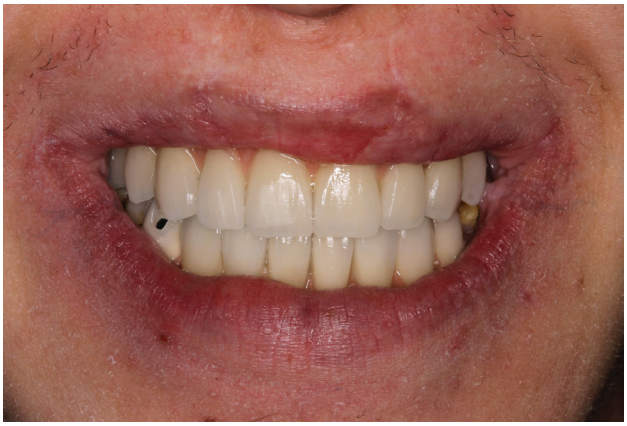

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FIG. 5 Clinical session for ceramic try-in of definite implant-supported fixed dentures. A. View with lip in rest position. B. View of the smile.

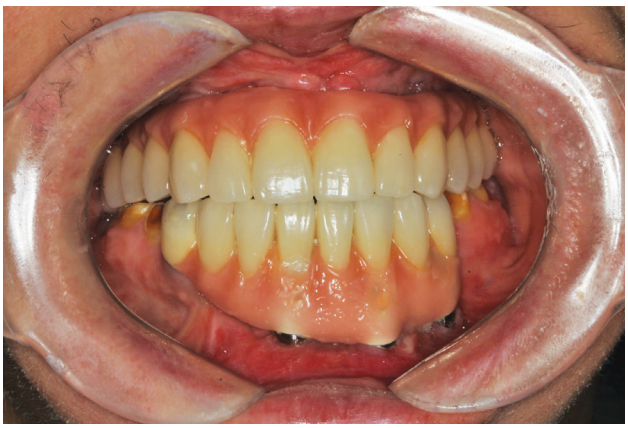

FIG. 6 Follow-up after two years of prostheses installation.

new prostheses and oral tissue remodeling under loading. Approximately one year after the temporary prostheses installation, the procedures for the definitive ceramic prostheses started. A new open tray impression and teeth assembly was made and tested in wax. A metal framework was cast in (Ni-Cr Beryllium free, Wironia Light, BEGO, Bremen, Germany) based on the teeth positioning and shape of the future ceramic coating (Fig. 4). The color A3 was selected for the dental cervical region and color A2 for the middle and incisal regions (Kuraray Noritake Dental Inc., Tokyo, Japan). The ceramic try-in was performed for aesthetic and occlusal adjustments, as well as for patient's approval (Fig. 5). After ceramic glaze, the maxillary and mandibular prostheses were installed with a 10 Ncm torque on the prosthetic screws. The mandibular screw holes were located on the gingiva area and were masked with a pink composite resin (Nexco Paste Gingiva 2 G2, Ivoclar Vivadent, Schaan, Liechtenstein) similar to gingival color. For other screw holes, an enamel composite resin color A2 was used (Charisma Classic A2, Heraeus Kulzer, Hanau, Germany).

The patient has been under follow-up every 6 months for clinical control. The patient adopted an oral hygiene protocol consisting in daily flossing under the bridge with Super Floss (Oral B, Procter & Gamble, Cincinnati, OH, USA), and twice-a-day brushing with a sulcus toothbrush for soft tissue-prosthesis sites, and with a traditional toothbrush for artificial teeth. After two years, all prostheses and implants showed clinical success, and the patient was very satisfied with the prosthetic treatment (Fig. 6).

DISCUSSION

This clinical report details the oral rehabilitation of a patient with ED and CLP using ceramic implant-fixed prostheses, with 2 years of follow-up. The case was challenging because of the patient's clinical history and local conditions, as well as his high expectations with the prosthetic treatment.

ED dental alterations present several limitations for retention and stability of conventional fixed or removable prostheses, since missing teeth and malformations make the dental preparation and use of intraradicular retainers very difficult (9, 14). The treatment of ED patients, and especially with CLP, starts at a very young age, almost immediately after birth, including surgical repairs of the cleft area and early bone grafts (10). Implant therapy for these patients follows a standard protocol as for individuals without ED or CLP, which is after growth completion and other procedures before prosthetic treatment, such as orthognathic surgery (3).

Even though the natural teeth were not adequate to support fixed prostheses, the surgical jaw repositioning was important to establish an acceptable anatomical relation, which was necessary to fabricate the prostheses in correct occlusion. After an improved skeletal position was established, the lip support was compensated with new prostheses. Without that procedure, the maxillary prosthesis would need a large vestibular compensation, leading to loss of lip support, eversion of upper lip when smiling, screw fracture, poor biomechanics, and risk for implant failure (16). However, some common sequelae of CLP still were present, such as maxilla atrophy, hypertrophic scar, and lip-notching (2, 6). Removable prostheses with dental retention systems have several positive features, such as oronasal communication sealing, proper lip support, easy hygiene access, and less invasive procedure with no need of surgery (7). However, the patient wanted to have fixed dentures and was willing to have implant surgery. Therefore, the chosen treatment plan was implant-supported fixed dentures after examining the adequate bone quantity and quality and assessing potential risk factors for dental implants (5). Because of the anatomic position of the mental foramen, the implants needed to be placed in a more anterior region, although the literature shows cases of successful prostheses with distal cantilevers (17). Considering the patient's conditions and the possibility of maintaining the second premolars and first molars, the safest option was chosen: an implant-fixed denture with reduced arch to minimize the cantilever that would have approximately two pre-molars and one molar each side. The presence of natural teeth in high masticatory force regions may prevent an unfavorable occlusal load distribution along the prosthesis distal portion and possible fractures (12,13).

As the patient always has reduced VDO due to maxillary atrophy (7), the use of temporary fixed prostheses allowed the adaptation to the new increased VDO established through functional and aesthetic clinical parameters. However, an exaggerated increase in vertical dimension can lead to a reduction in interocclusal space, elongated face, temporomandibular joint alteration, and occlusal interferences during phonation and masticatory function. Patients usually describe a feeling of "oral claustrophobia" for not having enough internal oral space with excessive VDO (11).

After the adaptation period with temporary acrylic prostheses in the established vertical dimension, the fabrication of the definitive prostheses in ceramic started. One of the reasons that metal-ceramic prostheses were chosen over conventional acrylic dentures was that ED patients usually have xerostomia (8). Reduced salivary flow combined with acrylic resin microporosity, which may act as a vessel for microorganisms (18), result in a suitable environment for accumulation and proliferation of bacteria and fungi (mostly Candida albicans), leading to an increased risk for mucosa inflammation (19). Moreover, ceramic teeth are very aesthetic and have less wear than acrylic teeth (23), but framework design planning is more complex and requires additional clinical and laboratorial steps (24). Besides lengthy treatment and high costs of oral rehabilitation with ceramic implant-supported prostheses, the results for patients with CLP and ED are very satisfactory (6,18 5, 14).

Clinical follow-up was established as a recall every three month initially, but a 6-month recall was adopted as a lifelong strategy due to the patient's commitment and good plaque control (20). The oral rehabilitation longevity strongly depends on the patient's commitment to home care, and the dental team should screen for possible alterations in soft or hard tissues, such as plaque and calculus accumulation, pain, bleeding on probing, implant/dental mobility, prosthesis and implant integrity, occlusion balance, and radiographic conditions of peri-implant structures.

CONCLUSION

Fixed and removable prostheses supported by teeth or implants can be used for prosthetic rehabilitation of CLP patients. However, ED often causes alterations in the shape and position of natural teeth, which prevent them being used as suitable retainers for fixed or removable dentures. This report shows that ceramic implant-supported fixed prostheses are predictably safe when sufficient alveolar bone is present in a patient with both CLP and ED, respecting the patient's desire to have a pleasant smile and the comfort of fixed dentures.

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Contributions

TGA and RSAS wrote the manuscript; TGA, RLOS, CMPS and RMHS were responsible for the clinical and laboratorial procedures; RSAS and MRLP developed the study protocol; and TGA was responsible for the overall data analysis/interpretation.

Conflicts of interests:

The authors declare no potential conflict of interests.

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