Fixed rehabilitation of a patient with hypohidrotic ectodermal dysplasia using zygomatic implants

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We found few references in the dental literature on implant rehabilitation of patients with hypohidrotic ectodermal dysplasia and only 1 case on zygomatic fixations and maxillary prosthesis in a patient with hypohidrotic ectodermal dysplasia. Two zygomatic fixations were placed according to the sinus slot technique, together with 3 implants in the anterior maxillary region. After 6 months, an upper complete prosthesis was screwed onto the implants, and lower overdentures were placed over the remnant canines. After 18 months of follow-up the patient reported important improvement in oral function and self-esteem. (Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2004;98:161-5)

Hypohidrotic ectodermal dysplasia (HED) linked to chromosome X is a hereditary disorder characterized by abnormal development of tissues derived from the ectoderm; it is observed in 7 out of every 100,000 live births.1 The most frequent oral manifestations are oligodontia or anodontia, delayed dental eruption, teeth shape alterations, enamel hypoplasia, and deficient development of the alveolar process.2-5 Early dental loss and alveolar osseous atrophy make conventional removable prosthetic adaptation difficult.1,3 The placement of endosseous implants for subsequent restoration is complicated, owing to the diminished bone volume available for the placement of implants,6 and bone grafting may be required.7

In 1998, Brånemark8 presented a zygomatic fixation as an alternative for rehabilitation of the atrophic maxilla. Stella and Warner9 posteriorly described the insertion of zygomatic implants using the sinus slot technique. This procedure consists of drilling a hole through the bone and into the sinus cavity in the upper area of the contour of the zygomatic buttress. A second bur hole is then made on this line 5 mm above the crest of the ridge, and a slot is made to connect the 2 holes. The ideal location of the implant abutment is on the crest in the first molar region. A drill is guided through the center of the slot and advanced superiorly towards the junction to the lateral orbital rim and zygomatic arch. The implant is placed following the slot under direct visualization, and a hexagonal screwdriver helps ensure proper angulation. This technique improves upon the original method in certain aspects, including the provision of a more vertical orientation of fixation, emergence over the alveolar ridge, and elimination of the sinus window.

The present study describes fixed rehabilitation of the maxilla in a patient with HED who presented maxillary anodontia, mandibular oligodontia, and severe atrophy of the residual alveolar crest margin. Maxillary rehabilitation was carried out over 2 zygomatic implants positioned according to the sinus slot technique, with 3 implants placed in the anterior zone.

CLINICAL CASE

A 29-year-old male presented hypohidrotic ectodermal dysplasia (HED). Extraoral examination revealed generalized hypotrichosis, frontal bulging, a depressed nasal bridge, lip thickening, and a prominent chin (Fig 1). The intraoral study in turn showed total upper (Fig 2) and partial inferior edentulism (Fig 3), severe maxillary and mandibular atrophy, and the presence of metal stumps cemented to remnant teeth 33 and 43—the latter having migrated towards the midline. The purpose of management was to solve the functional and esthetic problems caused by the lack of teeth—a situation that had not been satisfactorily resolved with conventional removable dentures. Orthopantomography and computed axial tomography (Fig 4) revealed extreme maxillary atrophy with little remnant bone. Based on these findings, we proposed maxillary fixed rehabilitation over implants (including 2 transzygomatic implants), with the placement of inferior overdentures on the remaining mandibular teeth.
Surgery was carried out under intravenous sedation with 1% propofol and vital functions monitoring by an anesthetist. Local anesthesia was administered (4% articaine with 1/100,000 epinephrine), and a supracrestal incision was made from one tuberosity to another, with distal extensions (Fig 5). The beds were prepared in the anterior zone with the help of osteodilators. Three ITI® SLA implants (Straumann, Walderburg, Switzerland) measuring 12 mm in length and 4.1 mm in diameter were positioned—2 in the remnant bone of the canine processes and 1 in the cortical component of the nasopalatal duct, this being the only location affording sufficient bone. We placed 2 Brånemark® system zygomatic implants (Nobel Biocare, Göteborg, Sweden) in the area of missing teeth 3 and 14, measuring 35 mm in length, by means of the sinus slot technique (Figs 6 and 7). Amoxicillin (750 mg) and ibuprofen (600 mg every 8 hours) were prescribed for 7 and 4 days, respectively. The postoperative evaluations were made after 7, 14, 30, and 60 days.

After 6 months, an upper complete prosthesis was screwed onto the implants, and lower overdentures were placed, supported by the metal stumps of 21 and 28 (Fig 8). After 18 months of follow-up the patient reported important improvements in oral function and self-esteem. Both the implants and prostheses appeared normal (Figs 9 and 10).
DISCUSSION

Our patient presented alterations of the skin, hair, sweat glands, teeth, and facial features characteristic of hypohidrotic ectodermal dysplasia (HED).\(^{10-12}\) He was edentulous in the upper jaw, with severe maxillary bone atrophy. In the dental literature, few studies have described prosthetic rehabilitation over implants in patients with this disease, and the cases that have been reported involved conventional implant techniques.\(^{3,6,11,13-16}\)

Fig 5. Intraoral image showing the extreme maxillary atrophy.

Fig 6. Zygomatic fixture on the prepared slot and perforated zygomatic-maxillary process.

Fig 7. Panoramic view showing the location of the zygomatic fixations and 3 implants mesial to them. The metal stumps on the canines can be seen in the mandible.

Fig 8. Image of both prostheses and their occlusal relation.

Fig 9. Occlusal view of the maxillary fixed prosthesis and implant emergence.
In ectodermal dysplasia the diminished bone volume available for placing implants, and the limited success of implants in the upper maxilla limits the possibilities of fixed rehabilitation; as a result, it is not possible to position implants with primary fixation using the conventional technique, as indicated by Mattsson et al. The limitations inherent to the use of bone grafts, or the existence of major defects in the atrophic upper jaw, have led to the search for alternative dental implant anchoring sites. We did not consider the use of bone grafts or sinus lifting, due to the increased complexity of the surgery involved and the lesser percentage success of implants positioned over grafted bone. In our case the considerable bone resorption of the jaw made primary implant placement impossible, and zygomatic fixtures were thus considered the best option.

Transzygomatic implantation is a novel technique. Aparicio and Malevez, based on a series of 29 clinical cases, described the characteristics of this approach in terms of its indications, surgical particularities and the clinical procedure for prosthesis elaboration. Parel et al. carried out a retrospective study of 65 zygomatic implants positioned in 27 patients (24 subjected to maxillectomy, and 3 with an alveolopalatal fissure); after an average follow-up of 6 years. None of the implants were lost. These implants must be combined with a minimum of 2 implants in the anterior sector and must be stent-fixed by means of a prosthetic superstructure.

We have found only one implant rehabilitation case similar to our own, published by Balshi and Wolfinger. These authors successfully treated a 20-year-old patient with ectodermal dysplasia by maxillary prosthetic rehabilitation over zygomatic implants and using the conventional Branemark technique. Our zygomatic implants were placed using the sinus slot technique, thereby avoiding the sinus window and bone loss. Moreover, our technique was less traumatic and afforded biomechanical advantages by orienting fixation in a more vertical direction with respect to the occlusal plane. In addition, 2 implants were placed in the canine area and a third in the nasopalatal duct—no references being found in the literature to insertion in this location.

Fixed rehabilitation of the maxilla over implants with inferior overdenture mandible placement not only afforded oral functional and esthetic improvements in our patient, but also improved patient self-esteem. In our patient with hypohidrotic ectodermal dysplasia, the use of zygomatic fixations placed according to the sinus slot technique constituted a valid rehabilitation alternative, though long-term evaluation is required.

REFERENCES

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