Oral rehabilitation with endosseous implants in a child with ectodermal dysplasia: a case report

Abstract

Aim The purpose of this article is to report the clinical course of a 12-year-old child with ectodermal dysplasia who was treated with an implant-supported overdenture for the mandible and an overdenture for the maxilla.

Case report Two dental implants were placed in the canine regions of the mandible. The maxillary teeth were prepared for the milled copings. Because the preparation of parallel walls was difficult, near-parallelism with an angle of convergence or taper of approximately 5 degrees was achieved. The cervical third of the teeth was prepared to be as parallel as possible to one another. In addition, the occlusal surfaces were reduced 1.5 mm, and the axial surfaces were reduced 1 mm. Occlusal reduction was performed to provide adequate thickness for the overlying denture base material. A chamfer finish line was prepared. The copings were cast with a Cr-Ni-based metal alloy and luted, bilateral balanced occlusion was developed using anatomic acrylic teeth. An impression was taken with an individual tray for impressions of overdentures. In response to the patient’s dry mucosa, the impressions were taken using rapid-setting silicone impression material with high elasticity. Bilateral balanced occlusion was achieved using anatomic acrylic teeth for overdentures. The maxillary overdenture and implant-supported mandibular overdenture were prepared by conventional methods using thermal-curing acrylic resin. The patient was seen 48 hours later for adjustment, then after 1 and 2 weeks, 1, 3, and 6 months, and 1 year and he is still satisfied with his prosthesis both aesthetically and functionally.

Conclusion The use of endosseous implants in the prosthetic rehabilitation of children with ectodermal dysplasia may provide a considerable improvement in comparison with traditional prosthetic methods.

Keywords: Child; Ectodermal dysplasia; Endosseous implant; Overdenture.

Introduction

Ectodermal dysplasia (ED) syndrome was first described in the medical literature by Thurnam, who reported two typical patients in 1848 [Nyhan, 1987]. The condition is classified into two major types: hypohidrotic, in which sweat glands are absent or significantly decreased; and hidrotic, in which sweat glands are normal [Guckes et al., 2002]. Hypohidrotic ectodermal dysplasia (HED) is characterized by hypohidrosis, hypotrichosis and hypodontia and occurs with a frequency of 1/100,000 births. It affects men severely, while women carriers and heterozygotes usually show minor defects: the complete syndrome, however, has also been reported in women [Nyhan, 1987; Neville et al., 1995]. The hidrotic form, inherited as an autosomal trait, does not ordinarily involve sweat glands [Gorlin et al., 2001]. The various forms involve dysplasia or aplasia of two or more ectodermal structures (skin, hair, nails, eyes, teeth, sweat glands, parts of the eye and the ear, sensorineural tissues, adrenal tissue) [Neville et al., 1995]. In general, the skin of the affected child is lightly pigmented and appears thin, with surface blood vessels easily visible. In most cases, the skin is dry, scaly and easily irritated as a result of poorly developed or absent sebaceous glands. Sculp hair, eyebrows, eyelashes and other body hair may be sparse, poorly developed or absent. Fingernails and toenails may also show faulty development [Levin, 1988; Neville et al., 1995].

Oral findings are often significant and can include multiple tooth anomalies (such as anodontia and hypodontia) and lack of normal alveolar ridge development [Levin, 1988]. In many cases HED exhibits the most severe dental anomalies, that require the assistance of the prosthodontist and orthodontist in the clinical management. Historically, prosthetic treatment for HED patients involved removable partial dentures, complete overlay dentures and fixed partial dentures [Yenisey et al., 2004]. Dental implants provide an additional treatment option for rehabilitation of these patients [Sweeney et al., 2005]. There are aesthetic, functional and psychological reasons that make it important to start oral rehabilitation early in life [Bergendal et al., 1991; Kramer et al., 2007]. The use of dental implants is a well-established treatment modality in adult patients who have ceased growth [Adell et al., 1990], and has also proved effective in certain situations in growing individuals including those with ectodermal dysplasia [Kearns et al., 1999; Guckes et al., 2002; Sweeney et al., 2005; Kramer et al., 2007].

The aim of this case report is to present a case of oral rehabilitation of the mandible with two dental implants in the canine region and maxilla with coping-retained overdenture in a 12-year-old patient with ED.

Case report

A 12-year-old boy with partial anodontia referred to the paediatric dental clinic because he had difficulty in chewing, and his prosthesis displaced when chewing and speaking. The patient’s dental history, obtained from the parents, indicated that the child’s current prosthesis had been done one year earlier and had been relined six
months before the visit. At the time of the visit, however, the child complained about the prosthesis. The patient had been diagnosed with HED when he was three years old, and displayed the typical characteristics of ectodermal dysplasia, such as brittle and fine blonde hair, a saddle or bulbous pear-shaped nose, a horizontal groove on the chin, thick cheeks and large ears (Fig. 1a, b).

Intraoral examination revealed a dry oral mucosa, protuberant lips, a thin vermilion line and undeveloped alveolar ridges. There were two cone-shaped, hypoplastic anterior teeth in the maxilla. Radiographic examination showed that the mandible was edentulous (Fig. 2). The patient exhibited loss of vertical dimension.

Clinical and radiographical examination revealed that excessive resorption and atrophy of the alveolar ridges had occurred. According to the clinical examination, his old mandibular prosthesis was not in harmony with the mandibular crest. A diagnostic cast was prepared and it was decided to apply an implant-supported overdenture for the mandible and a coping-retained overdenture for the maxilla. Based on the need to increase the stability and retention of the mandibular denture, the decision was made to apply an implant-supported prosthesis. As a result, the patient's speech, chewing, swallowing and aesthetics noticeably improved. Also, it is anticipated that the patient's quality of life and social integration will improve.

Before implant surgery, mandibular alveolar height was detected with computerised axial tomography. Implant sizes and locations were defined with Stend cat programme (Fig. 3). Endosseous implant treatment protocol was planned. Two dental implants (Strauman Standard Plus Implant, Strauman, Switzerland) (diameter 3.3 mm, length 10 mm) were placed in the canine regions of the mandible under local anaesthesia. Postoperative clinical and radiographic evaluations of the inserted implants were made after 7, 30, 60 and 90 days (Fig. 4). After a submerged healing period of three months, the implants were exposed, and abutment connection was performed. The patient was prescribed penicillin 1000 mg 2x1 for seven days and healing was uneventful. On postoperative day two, oral hygiene instruction was given and the patient was prescribed 0.2% chlorhexidine mouthwash for 10 days. Prosthodontic procedures were started as soon as the soft tissues around the abutment cylinder had healed (Fig. 5).

In the evaluation, the coronal structures of the maxillary teeth, axial wall inclination and relative positions were considered to determine the path of insertion, retention and stabilization of the overdenture, to minimize plaque accumulation on the hypoplastic teeth, and to optimize the crown/root ratio of the fabricated metal copings. While preparing the teeth for the metal copings, the slopes of the axial walls were made nearly parallel. The teeth were prepared for the milled copings. Because the preparation of parallel walls was difficult, near-parallelism with an angle of convergence or taper of approximately 5 degrees was achieved. The cervical thirds of the teeth were prepared to be as parallel as possible to one another. In addition, the occlusal surfaces were reduced 1.5 mm, and the axial surfaces were reduced 1 mm. Occlusal reduction was performed to provide adequate thickness for overlying denture base material. A chamfer finish line was prepared. After the teeth were prepared, a definitive impression of the dome-shaped teeth for coping fabrication was made with a custom tray using medium-viscosity silicone impression material (Elite H-D Medium,
Zhermack, Rovigo, Italy). The copings were casted with a Cr-Ni-based metal alloy (Wiron 99, Bego, Germany) and cemented to the teeth using polycarboxylate cement (Adhesor Carbofine, SpofaDental, Czech). An impression was taken with a custom tray for impressions of overdentures, individual acrylic trays were fabricated, then bolder molding was performed using impression compound (Kerr, Salerno, Italy). In response to the patient's dry mucosa, the impressions were taken using rapid-setting silicone impression material with high elasticity [Anusavice, 1996; O'Brien, 1997]. First the dimension of the rest position was measured, then the vertical dimension of occlusion was determined, approximately 3 mm above the rest dimension. To determine the previous vertical dimension of occlusion, the maxillo-mandibular relationship of the patient was recorded using occlusion rings in centric relation. Bilateral balanced occlusion was developed using anatomic (cusp angle of 33°) acrylic teeth. The maxillary overdenture and implant-supported mandibular overdenture were prepared by conventional methods using thermal-curing acrylic resin (Figure 6). The patient was seen 48 hours later for adjustment, then at one, two weeks, one, three, and six months and one year. He is satisfied with his prosthesis both aesthetically and functionally.

Discussion

Severe hypodontia or even anodontia in children are very rare conditions, most often associated with congenital syndromes such as ED [Silverman and Ackerman, 1979]. Prosthetic rehabilitation of these patients is a routine protocol. Endosseous implants can be considered an alternative treatment [Guckes et al. 1991; Kearns et al., 1999; Sweeney et al., 2005]. But in edentulous children who have ED, multiple implant placement is not possible because of the ongoing development of the jaws and insufficient bone volume and height. As a routine practice, the placement of implants in growing children is not recommended [Guckes et al. 1991; Cronin and Oesterle, 1998; Kearns et al., 1999]. This is because growing implant patients present a unique age-related problem with regard to implant positioning and prosthetic outcomes [Bergendal et al., 1991; Bergendal et al., 1996]. This is especially critical in the maxilla, where implants can be submerged by the downward growth of developing tissues. Implants also can become submerged relative to adjacent natural teeth as growth occurs. However, as the lateral growth of the anterior mandible is usually completed by three years of age, if anodontia exists in this area, implant placement can be considered [Escobar and Epker, 1998]. In this case, the patient could not use his removable prosthesis and he was not satisfied with it, neither aesthetically nor functionally. The dental implants were placed in this patient only in the anterior region of the mandible where implants have a lower risk of submersion because of less vertical bone growth [Odman et al., 1991; Ledermann et al., 1993]. An implant-supported overdenture for the mandible and an overdenture for the maxilla were also applied. The purpose of management was to solve the functional and aesthetic problems caused by the lack of teeth and alveolar process.

In anodontia cases and especially in HED, growth of the alveolar bone is seriously impaired [Yenisey et al., 2004]. However, the functional stimulation transferred by a removable prosthesis may help growth of the ridge. In this case, it may also be necessary to adapt and change some of the mandibular prostheses to the modification in occlusion. For this reason, the ball-retained overdenture may be a more practical prosthesis for rehabilitation of patients whose growth is not complete. In spite of the successful integration and survival of the implants in all age groups, practical considerations (including cost/benefit ratio) mandate that the timing of implant placement be considered carefully for each individual patient. Most preadolescent patients adapt to conventional removable prostheses. There is little evidence that the use of dental implants to orally rehabilitate prepubertal patients with multiple congenitally missing teeth will positively affect the growth and development of the craniofacial complex [Guckes et al., 2002].

Conclusion

For selected cases and certain sites, implants can provide a useful alternative to conventional treatment and provide the support and retention benefits of an overdenture.

References

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