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A prosthetic treatment approach for a cherubism patient: A clinical report

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Cherubism is an early childhood disease that primarily involves the mandible and consists of painless mandibular enlargement with or without maxillary involvement and progresses rapidly over the course of several years. This clinical report describes the fabrication of maxillary fixed partial dentures and a mandibular overdenture for a 21-year-old man with cherubism. (J Prosthet Dent 2006;96:313-6.)

Cherubism is a rare, inherited developmental abnormality that causes bilateral enlargement of the maxilla and/or mandible.[1] This disease is caused by an autosomal dominant gene located on chromosome 4p16.3,[2] and typically affects men.[3] It is generally accepted that it is a benign disease of bone, beginning at the age of 2 or 3 years, that progresses in childhood, with a peak at the age of 5, and shows spontaneous regression at the end of adolescence.[3] Jones[4] was the first to describe the disease in a family and suggested the name. The findings in his original patients as adults,[5] and in their children,[6] were published in 1952 and 1965, respectively. The diagnosis of cherubism is based on clinical, radiographic, and histological findings.[3] The mandibular angle, ascending ramus, retromolar region, and posterior maxilla are most often affected. The coronoid process can be involved, but the condyles are always spared.[7]

The most common physical manifestation is a painless, firm, bilateral enlargement of the lower face. Enlargement of the submandibular lymph nodes may occur, but no systemic abnormalities are involved.[1] The clinical appearance may vary from barely discernible posterior swelling of the maxilla or mandible to a marked anterior and posterior expansion of both the maxilla and mandible, resulting in masticatory, speech, and swallowing difficulties.[7] Maxillary involvement usually results in the greatest deformity.[7] Maxillary expansion may produce the characteristic "eyes raised to heaven"[4,8] appearance, in which a thin band of sclera is exposed between the iris and lower eyelid.[8,9] The disease primarily occurs in the mandible, and Meng et al[10] reported in a long-term study that 19 of 24 cherubism patients had only mandibular involvement. Of the 5 with maxillary involvement, 1 had orbital expansion.

Radiographically, the lesions appear as multiple, well-defined, multilocular, radiolucent areas in the mandible and maxilla. These lesions begin in the posterior alveolar region and/or ramus and can spread anteriorly. The lesions are irregular in size and usually cause marked destruction of the alveolar bone. Numerous displaced and unerupted teeth appear to be floating in radiolucent spaces.[11] An occlusal radiograph of the maxilla may appear as soap bubble–like, with maxillary antrum obliteration.[7]

The reported treatment of cherubism has varied considerably and includes no active treatment, extraction of teeth in the areas involved, surgical contouring of expanded lesions, and complete curettage.[11] Treatment can be delayed, because the cystic lesions usually become static and fill in with granular bone during adolescence and at the end of the skeletal growth period.[1] There are usually no systematic manifestations, and laboratory values are usually within normal limits.[9] Long-term clinical studies have revealed that the childhood lesions give way to partial or complete resolution in the adult.[3,10,11] Radiation therapy is contraindicated,[9] and Mangion et al[2] reported an osteosarcoma in an irradiated area. The purpose of this clinical report is to present prosthetic treatment of a 21-year-old man with cherubism.

CLINICAL REPORT

A 21-year-old white man with a history of cherubism disease was referred by a general dentist to the Department of Prosthodontics, Ankara University. The patient's physical appearance showed characteristic cherubic features, including stretching of the skin of the cheeks and a bilaterally swollen posterior mandible and maxilla (Fig. 1). Difficulty with speaking was noted. Widespread lesions in the maxilla and mandible were identified on a panoramic radiograph (Fig. 2). Although lesions reported for cherubism patients are primarily detected in the mandible, the patient showed a greater presence of lesions in maxillary bone within the infraorbital area.

The maxillary canines and second and third molars were missing, with a diastema present between the maxillary central incisors. The lesions were so widespread in the mandible that all the teeth, except for the right canine and first and second premolars, and the left first and second premolars, were extracted before the patient was referred for treatment. The patient expressed concern about the position of his maxillary anterior teeth and inefficient masticatory performance. The patient's

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Fig. 1. Facial view of patient.

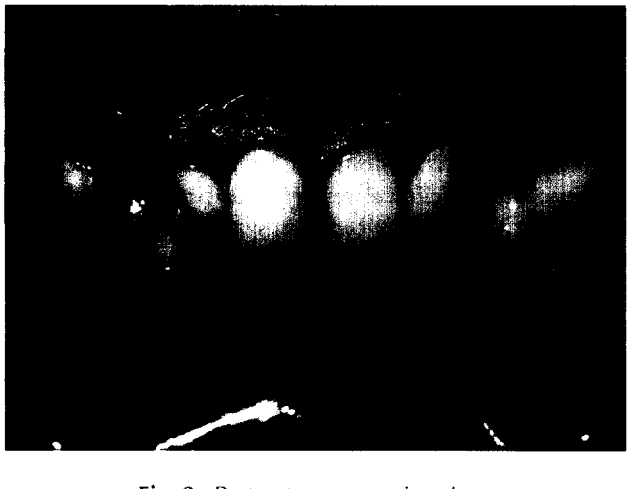

Fig. 3. Pretreatment anterior view.

concerns and expectations were considered in developing the treatment plan.

Maxillary and mandibular complete-arch impressions were made using irreversible hydrocolloid impression material (Cavex CA37; Cavex Holland BV, Haarlem, The Netherlands). Diagnostic casts were fabricated from type III dental stone (Moldano; Heraeus Kulzer GmbH, Hanau, Germany) and mounted in a semi-adjustable articulator (Hanau H2; Teledyne Hanau, Buffalo, NY) using face-bow transfer and a centric relation record.

Implant placement was initially considered for restoration of the edentulous alveolar spaces in the maxilla and mandible. However, the authors were concerned that

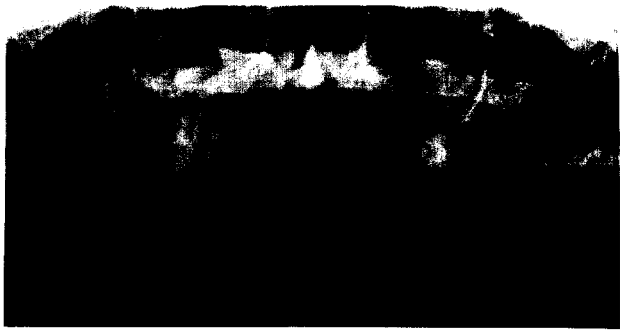

Fig. 2. Panoramic radiograph with multilocular radiolucencies.

implant placement into porous bone might result in lack of osseointegration and complications. Consequently, implant placement was not deemed to be a treatment option for either the maxilla or mandible. The number of remaining abutment teeth in the maxilla indicated that fixed partial dentures (FPDs) might be an option. For the mandible, alternative treatment options were a removable partial denture (RPD) or overdenture. A bilateral reverse articulation was noted due to the greater width of the maxillary dental arch (Fig. 3). Interdigitation of the existing teeth with an RPD in the mandible was not possible due to the disharmony of the arches. Thus, the treatment plan for the mandible included coverage of the mandibular teeth with copings, and restoration of the edentulous spaces with an overdenture.

As the treatment plan consisted of FPDs for the maxilla and an overdenture with copings for the mandible, maxillary and mandibular teeth were prepared using a diamond rotary cutting instrument (Medin; Nove Mesto na Morave, Czech Republic). Definitive impressions of the prepared maxillary and mandibular teeth were made using a vinyl polysiloxane impression material (Speedex; Coltène/Whaledent, Cuyahoga Falls, Ohio). Working casts were generated from type IV die stone (BegoStone Plus; BEGO, Bremen, Germany) and mounted in an articulator with face-bow transfer using interocclusal records.

Precision attachments offer considerable advantages in flexibility for treatment planning,[12] and they are used as retentive elements for FPDs, RPDs, and overdentures.[13] Resorption of the alveolar crest and insufficient abutment teeth for support in the patient's mandible impeded retention of the denture. Precision attachments seemed suitable to provide additional retention to the proposed overdenture. For this reason, precision attachments (Bredent, Senden/Witzighausen, Germany) were placed on the buccal surfaces of the mandibular copings with a surveyor (Cruise 440; Silfradent, S. Sofia, Italy) before the wax patterns of the splinted copings were cast. Parallel guide planes on the proximal and lingual surfaces of the abutment teeth or pontics can have a positive

Fig. 4. Maxillary fixed partial dentures and mandibular copings in place.

Fig. 6. Intraoral view of definitive prostheses.

effect on the stability and retention of an RPD, as might be expected with the use of precision attachments.[14] Stabilization grooves were prepared in the lingual surfaces of each coping with parallel guide planes on the proximal surfaces, with the aim of neutralizing the torquing effect of the attachments on the abutment teeth. Two maxillary FPDs were fabricated between the right central incisor and first molar, and the left central incisor and first molar, to minimize the diastema between maxillary right and left central incisors. The maxillary FPDs and the mandibular copings were cast with a Co-Cr alloy (Wirobond C; BEGO). The framework of the maxillary FPDs and the polished copings were evaluated intraorally for fit, retention, and marginal integrity. The mandibular copings were not cemented to make an impression of the overdenture. The definitive impression for framework of the overdenture was made with an irreversible hydrocolloid material (Cavex CA37; Cavex Holland BV) and a custom tray. The framework of the mandibular overdenture was cast from a cobalt-chrome alloy (Biosil F; Degudent GmbH, Hanau, Germany) and evaluated intraorally for fit, retention, and stability. The porcelain (Ceramco II; Dentsply Ceramco, Burlington, NJ) maxillary FPDs were evaluated intraorally, and an occlusion rim on the mandibular framework was used to record

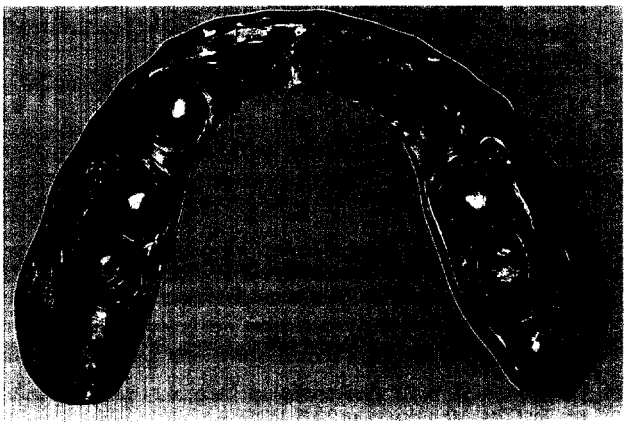

Fig. 5. Intaglio surface of mandibular overdenture with copings in place.

the maxillomandibular relationship. The casts were mounted in the same semi-adjustable articulator.

The mandibular artificial teeth (Vita Physiodens; Vita Zahnfabrik, Bad Sackingen, Germany) were arranged to direct forces to the buccal shelf area.[15,16] A bilaterally balanced occlusal scheme was developed. Trial placement of tooth arrangement of the mandibular overdenture was performed with the maxillary FPDs in place. The mandibular overdenture was processed with a heat-polymerizing acrylic resin (Paladent 20; Heraeus Kulzer GmbH), and the porcelain maxillary FPDs were glazed. Final occlusal adjustments of the overdenture were performed, and the overdenture was mounted on the copings. The copings and the glazed maxillary FPDs were cemented with zinc phosphate cement (Adhesor; SpofaDental, Prague, Czech Republic) (Figs. 4-6). The patient was instructed regarding care of the mandibular overdenture and maxillary FPDs and was placed on 6-month recall. The patient remained satisfied with the function and esthetic result at the 1-year recall appointment.

SUMMARY

Patients with cherubism may present with discrepancies in maxillomandibular relationships, irregular bone structure, and a lack of retention or stabilization for removable dentures. As presented in this clinical report, the use of implants may be restricted in these patients, and therefore, an overdenture may represent an alternative treatment option.

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0022-3913/$32.00

doi:10.1016/j.prosdent.2006.09.014