Functional and Esthetic Rehabilitation After Mandibular Resection in a Child Using a Tooth/Implant–Supported Distraction Device: A Case Report

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After resection and autogenous bone grafting in infancy and childhood, hypotrophy of the concerned jaw can often be observed. A 6-year-old male patient with osteogenic sarcoma was treated with partial resection of the mandible from the left first molar to the right first premolar. The aim was to rehabilitate the patient functionally and esthetically as he grew. At the age of 16, after the placement of 4 Brånemark System implants in the consolidated autogeneous bone graft, the patient was treated with horizontal distraction osteogenesis. A new and unconventional type of individual tooth/implant–supported distraction device was used to lengthen the mandible by 16 mm. The implants placed to support the device were later used for prosthodontic rehabilitation. Progress in bone reconstruction, plastic coverage, and implant dentistry, as well as distraction osteogenesis, have enabled the compensation of functional and esthetic impairments caused by resection, especially in infants and young children. INT J ORAL MAXILLOFAC IMPLANTS 2004;19:603–608

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Jaw resections in infancy normally lead to an underdevelopment of the affected jaw after the autologous osteoplasty. Facial deformations, reduced oral cavity size and access, innervation disturbances, and missing teeth often cause complications with masticatory function, deglutition, and speech mechanism.[1] The absence of functional stimulation of the bone graft by an appropriate force initiation represents another difficulty. The restoration of a congruent jaw position is necessary to avoid atrophy of the affected jaw due to disuse. At the same time, the orthodontic options available can be limited. Therefore, many cases require interdisciplinary cooperation and planning and unconventional and individualized methods of treatment.

Conventional surgical therapy for mandibular hypoplasia involves an operation described by Trauner and Obwegeser[2] or storing an autologous bone graft. However, if there is a significant bone deficiency, these surgical treatments often meet with difficulty because they can lead to soft tissue deficit or mucosal dehiscence,[3–5] a high recurrence rate caused by muscle and scar pulls,[2] transplant resorptions,[3,6] and morbidity to the donor.[3,7] Compared with these complications, callus distraction permits the evening of bone and soft tissue deficits to the same extent without the necessity of reverting to an autologous bone graft.[4,5] Therefore, callus distraction is particularly indicated for the lengthening of jaw areas malformed because of hereditary factors,[4,5,8] for the repositioning of jaw sections in patients suffering from dysgnathia,[5,9,10] for the closing of large bone defects,[11] and for the augmentation of atrophic sections.[6,10,12,13] When applying the conventional distraction osteogenesis procedure, the distraction mechanism needs to be fastened to the bone

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Fig 1 *(Above)* Site of lesion and operation.

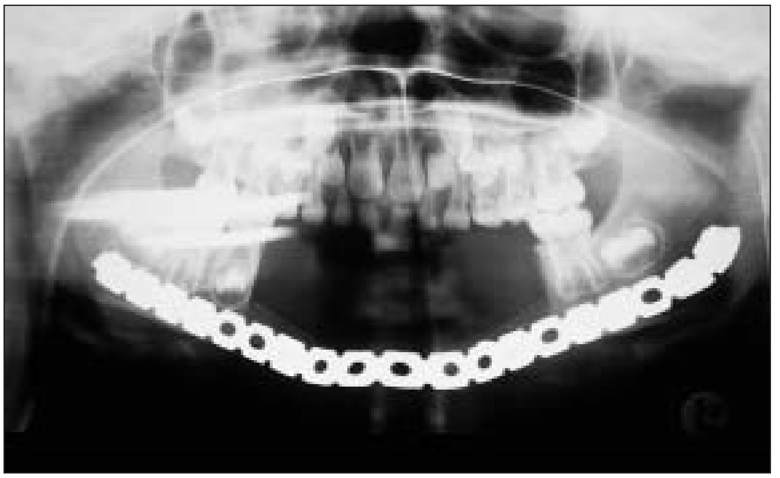

Fig 2 *(Right)* Panoramic radiograph after partial resection of the mandible from the left first molar to the right first premolar.

intraorally and submucosally and must be activated by a screw cylinder in the vestibule.

Michieli and Miotti[14] developed the tooth-supported distraction device for mandibular lengthening without osseous fixation in a veterinary experimental study in 1977. The success of this treatment has been confirmed by Niederhagen and associates[15,16] as well as Block and colleagues.[17,18] No serious destructive effects on the roots or the periodontium of the teeth in the proximity of the distraction gap were detected either clinically or histologically.[14,17,19] This method of fixation involves less risk of infection than conventional distraction osteogenesis because the oral mucosa is not penetrated. The control of progression in the phase of consolidation is less difficult, and the distraction device can be removed without surgery.[14]

In the present case, interdisciplinary treatment planning was conducted. After primary osteoplasty, both jaws were developed orthodontically as they grew. Then, when the conditions were favorable, implants were placed. These implants were first used to support the tooth/implant–supported distraction device for distraction osteogenesis and were used subsequently for prosthetic management.

CASE REPORT

The patient presented for the first time at the age of 6 years at the Clinic for Oral and Maxillofacial Surgery of the University of Bonn in 1991. The mandible was initially diagnosed as having an ossifying fibroma in the region of the mandibular left lateral incisor (Fig 1). However, after biopsy and histologic examination, the diagnosis was changed to osteoblastic osteosarcoma. Subsequently a segmental mandibulectomy from the region of the mandibular left second premolar to the mandibular right first premolar was performed. Both mandibular central incisors, the mandibular left second premolar, and the dental germs of the mandibular left and right second incisors had to be excised. The defect was reconstructed by means of primary osteoplasty (ie, a rib transplant) (Fig 2). Three months later the osteosynthesis plate was removed. At that time the rib transplant had not completely consolidated osteally. Due to pseudoarthrosis in the transitional zone, bone from the sixth and seventh ribs was stored and fixed by an osteosythesis plate and 3 perimandibular wires on March 25, 1993. The miniplates and wire ligature were removed 4 months later. The reconstructed mandible appeared to be completely osteally consolidated at this time. On July 12, 1993, the adjunctive orthodontic treatment was begun. The patient needed treatment for the following:

- An anterior diastema
- The protrusion position of the maxillary central incisors
- The “scissors bite” of the maxillary and mandibular right second primary posterior teeth and first molars
- The distoclusion of half a premolar (Fig 3)

A maxillary bite plate to close the anterior diastema and retrude the central incisors and a mandibular expansion plate to widen the posterior dentulous area were incorporated.

To gain height in the area of the rib transplants, augmentation using iliac crest bone fixated by titanium mesh was performed at the end of 1999. To make distraction osteogenesis and chewing function practical, it was decided to rehabilitate the patient with an implant-supported prosthesis. Four Brånemark System (Nobel Biocare, Göteborg,

Fig 3 *(Left)* Profile view of the 8-year-old patient postoperatively.

Fig 4 *(Below)* Status after exposure of 4 Brånemark System implants in the mandibular left lateral incisor, left first premolar, right central incisor, and right canine regions.

Fig 5a *(Left)* Profile view of the patient at 15 years of age.

Fig 5b *(Below)* Teleradiograph obtained before distraction osteogenesis.

Sweden) implants were placed in the grafted area on September 28, 2000—one in the region of the mandibular left lateral incisor, 1 in the region of the mandibular left first premolar, 1 in the region of the mandibular right central incisor, and 1 in the region of the mandibular right canine. The implants were exposed 6 months after placement (Fig 4). A cephalometric radiograph of the 15-year-old patient revealed the mandibular hypoplasia that gave him a bird-like face (Figs 5a and 5b).

A tooth/implant–supported distraction device with lingually situated screws was then constructed and placed on April 26, 2001. The device was anchored to the mandibular left first and second molars, the mandibular right second premolar, and the 4 implants. The appliance was produced individually by model casting (Remanium GM 800; Dentaurum, Ispringen, Germany) and fixed by screwed-on cast copings to the teeth and by individual gold abutments (UCLA Abutment; 3i/Implant Innovations, Palm Beach Gardens, FL) attached to the cast framework by laser. The device was activated by 2 screws (Hyrax Schrauben, Forestadent, Pforzheim, Germany) that were turned periodically. The cast copings were cemented on the teeth by glass-ionomer cement (Ketac-Zem; ESPE, Seefeld, Germany) (Fig 6a). To disengage the occlusion a maxillary bite plate was made. Subsequently an extraoral bilateral osteotomy was carried out in the regions of the mandibular left second premolar and right first premolar, and a test activation of 1 mm was performed. After a latency period of 7 days, the appliance was activated 1 mm per day over a period of 16 days (from May 3, 2001 to May 18, 2001) to elongate the mandible 16 mm

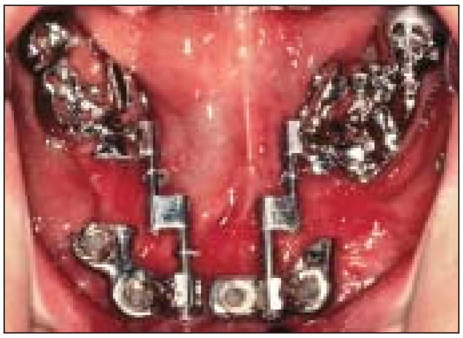

Fig 6a *(Left)* Distraction system after lengthening the mandible by 16 mm.

Fig 6b *(Bottom left)* Panoramic radiograph obtained after lengthening the mandible by 16 mm.

Fig 6c *(Bottom right)* Teleradiograph obtained after distraction osteogenesis.

(Fig 6b). On July 7, 2001, the distractor was removed after a consolidation period of 74 days (Fig 6c). Subsequently, to improve the soft tissue situation, the oral floor and vestibule from the region of the left first premolar to region of the right first premolar was grafted using free skin from one of the patient's legs.

Further prosthetic treatment was provided after a consolidation period of 4 months. The mandible was restored with a fixed prosthesis. Splinted ceramic-veneered crowns were seated on the left second premolar and first molar and the right first and second molars using mesial, individually molded attachments for each side. Anchorage of the implant-supported prosthesis was achieved using individually produced mesostructures (Fig 7a). The ceramic-veneered reconstruction was incorporated provisionally (Fig 7b).

RESULTS

As the result of cooperation between the departments of Oral and Maxillofacial Surgery, Orthodontics, and Prosthetic Dentistry, a functionally and esthetically satisfying rehabilitation of this patient could be achieved, despite extreme hypoplasia of the mandible after jaw resection and primarily osseous reconstruction in childhood (Fig 7c).

The distraction osteogenesis that was performed required a horizontal shift of the position of the autologous bone graft of approximately 16 mm. The desired bilateral lengthening of the mandible could be achieved by a distraction of 1 mm per day for 16 days. The improved maxillomandibular relationship made further prosthetic treatment of the patient possible. Seven months elapsed between the insertion of the distraction device and the loading of the definitive prosthesis. The attached gingiva did not show wound dehiscence at any time. Neither the teeth nor the implants demonstrated any loosening after the distraction.

Prosthetically, the young patient could be rehabilitated both functionally and esthetically with a fixed prothesis. The improved maxillomandibular jaw relationship after callus distraction ensured improved occlusion and lessened the risk of peri-implant bone graft resorption. In addition, stable support and positioning of the mandible prevented unwanted tooth migration.

DISCUSSION

Interdisciplinary Cooperation

Interdisciplinary treatment is very important in facilitating predictable results in cases of extensive

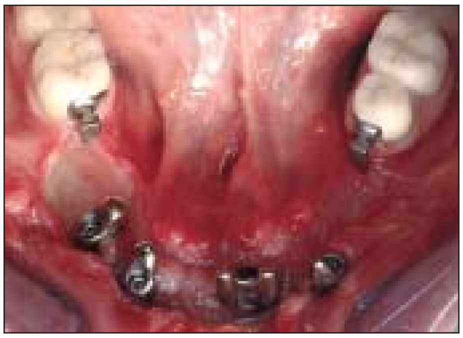

Fig 7a *(Above left)* The implants are shown with individually produced mesostructures. Splinted, ceramic-veneered crowns are seen on the right second premolar, left first molar, and left second molar, with mesial, individually molded attachments.

Fig 7b *(Above right)* The incorporated prosthesis extending from the left second molar to the right first molar.

Fig 7c *(Right)* Profile view of the rehabilitated patient.

rehabilitation. The development of the alveolar arch of the maxilla and correction of tooth position by orthodontic means occurred according to projected guidelines, since for a long period of time there was only a partially edentulous and extremely underdeveloped mandible. Therefore, development of the mandible was restricted to the dentulous jaw area. Here the posterior segment was widened transversally so that the existing "scissors bite" caused by the hypoplasia could be repaired. However, with the completion of these measures orthodontic treatment had reached its limits, and further surgery was required.

The success of traditional jaw surgery in the form of mandibular advancement after Trauner and Obwegeser[2] can be regarded as highly uncertain with a bone and soft tissue deficit of 16 mm. In extreme cases such as the present one, individually conceived measures of treatment are necessary. For that reason the planned fixed prosthetic care for the patient had to be carried out from 2 perspectives: the positioning of the implants had to be ingeniously planned to facilitate callus distraction and yet be efficiently positioned for prosthetic treatment after callus distraction.

Peculiarities of the distraction system

Michieli and Miotti,[14] as well as Block and coworkers,[17,18] presented veterinary experimental studies that proved that only tooth-supported distraction osteogenesis for mandibular lengthening without bony fixation was possible. The success of this method was confirmed and successfully applied to patients by Niederhagen and Braumann.[15,16] By means of this distraction method, sufficient stability and movement of the gnathic parts could be achieved. Neither destructive alterations in the root area or the periodontium of the teeth nor looseness of the implants could be observed either clinically or histologically.[15,16,19] The force needed after sagittal osteotomy for mandibular lengthening in human beings is 14 N; this force is proportional to the resistance of the surrounding structures (muscles, connective tissues).[20] In comparison, the force required for brisk maxillary expansion in monkeys (*Macaca fascicularis*) is 20 N, which is greater than the force essential for a callus distraction.[21] Thus, loosening of teeth or implants placed in tissue created by callus distraction is highly unlikely. In orthodontics, implants are considered inflexible retention elements of orthodontic appliances for tooth movement over longer periods of time.[20,21] Therefore, implants are especially suitable as retention elements for appliances for distraction osteogenesis. Seven months after callus distraction, the implants could be rated –1 to 0.5 according to the firmness test with the Periotest (Siemens, Bensheim, Germany). The results revealed that they were clinically firm.

Tooth/implant–supported distraction osteogenesis has a number of advantages compared with conventional distraction osteogenesis, in which the device is intraorally, submucosally fixed to the bone and activated by a screw cylinder in the vestibule. Penetration of the oral mucosa, which might increase the risk of infection, can be avoided; progression control during the time of consolidation is made easier; and the distraction device can be removed without surgery.[14] However, the relatively large, intraorally positioned tooth/implant–supported distraction device constricts the patient's mouth.

In principle, osteotomy of the mandible always involves the risk of injuring the inferior alveolar nerve. Because of the segmental mandibulectomy that preceded the osteotomy, there was no risk of nerve lesion in this case. Extraoral access to mandibular osteotomy was chosen because of unfavorable mucosal conditions in the vestibule caused by repeated surgical interventions; the patient already had a number of extraoral scars, so scarring unblemished tissue was not an issue. Furthermore, it would have been necessary to wait for complete soft tissue recovery before the distraction to prevent wound dehiscence caused by the extension. Beyond this, the waiting time for healing of the intraoral soft tissue can be regarded as a risk of a premature ossification of the osteotomy gap.

Prosthetic Treatment

As an alternative to conventional prosthetic treatment, several single prostheses or telescopically retained removable prostheses could have been used. However, the present method was designed to achieve primary splinting of the implants with the remaining teeth to stabilize the mandible. A loosening of the restoration in the area of the tooth/implant–supported crowns by elastic deformation of the mandible, especially around the molars, should be reduced by bilateral attachment connections. Temporary cementation of the prosthesis was selected to facilitate corrective plastic surgery. Likewise, necessary changes in the prosthesis could also be made in the area of the adjacent soft parts or the occlusion.

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