Implant-Supported Rehabilitation of an Edentate Patient with Osteogenesis Imperfecta: A Case Report

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This is a review of the literature on osteogenesis imperfecta and a case report of an edentulous patient with osteogenesis imperfecta rehabilitated with implant-supported fixed prostheses in the maxilla and mandible. Quality and quantity of the bone is of paramount importance for establishment of osseointegration. In osteogenesis imperfecta bone is osteoporotic. There are few reported cases in the literature of implant placement and subsequent rehabilitation of patients with osteogenesis imperfecta. To our knowledge, this is the first reporting of successful short-term follow-up of an edentulous osteogenesis imperfecta patient with implant-supported fixed prostheses. Int J Oral Maxillofac Implants 2008;23:947–952

Key words: *dental implants, dentinogenesis imperfecta, edentulous, osteogenesis imperfecta, sinus augmentation*

Osteogenesis imperfecta (OI) is a heterogenous group of bone disorders resulting in generalized osteoporosis of the skeleton.[1] In this genetic osteoporosis, the underlying defect is in collagen synthesis and specifically type I collagen.[2]

Because around half of the collagen in the body is in the skeleton, bony abnormalities in these patients are the most striking. Long bones have a thin cortex composed of immature woven bone. There is decreased volume and poorer quality medullary bone, and lamellar bone is more cellular in nature. Thus, bones are more fragile and susceptible to fracture.[3] They may also heal with excessive callus formation. Hence, progressive deformity due to multiple fractures is a prominent feature. Long bones are more commonly affected, although the skull can be affected. If the jaws are involved, OI is more commonly associated with the mandible.

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Many other collagen-containing parts of the body may be affected.[4] In some cases, the eyes have a blue sclera due its thinness and underlying uveal pigment. There can be deafness due to distortion of the ossicles. Joints may show hypermobility because of lax ligaments. The skin may be more translucent in nature. Cardiac involvement may include defective heart valves. Some forms have an association with dentinogenesis imperfecta (DI).[5]

Following advances in genetics it became apparent that most cases of osteogenesis imperfecta are inherited in an autosomal dominant pattern. A classification based on this and subsequent phenotypes was proposed (Table 1).[6] Further types of OI have been described more recently[7]; namely, types V, VI, and VII. These have very distinct clinical, radiographic, and histologic features. They are not, however, associated with dentinogenesis imperfecta.

The features of dentinogenesis imperfecta have been described.[8] The teeth appear brown or blue in color with opalescence. This is present from the time of eruption and is due to disorganization of the structure of the underlying dentin. There is a reduction in the number of tubules, and the remaining tubules are wider than normal. Histologically, the mantle dentin layer is normal but there follows an irregular dentin structure with exaggerated laminated structure. The amelodentinal junction is thought to be normal in structure.[9] The enamel is subject to fracture very soon after eruption, which exposes the defective dentin to the oral environ-

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Table 1 Classification of Osteogenesis Imperfecta Associated with Dentinogenesis Imperfecta[2]

Type	Inheritance	Dentition	Bone features	Other features
IA	Autosomal dominant	Normal	Variable fragility Moderate deformity	Blue sclera 20% scoliosis/kyphosis
IB	Autosomal dominant	Dentinogenesis imperfecta	NA	NA
II	Autosomal dominant	Unknown	Very severe fragility Multiple fractures	Blue sclera
III	Autosomal dominant	Dentinogenesis imperfecta	Severe fragility Progressive deformity	White sclera (in adults) Kyphoscoliosis
IVA	Autosomal dominant	Normal	Moderate fragility Moderate deformity	White sclera Kyphoscoliosis
IVB	Autosomal dominant	Dentinogenesis imperfecta	NA	NA

ment. Consequently, the teeth are subject to rapid attrition, often as far as the gingival level.[10] There is constriction of the cervical portion of the tooth. The pulp chambers and root canals are often obliterated with abnormal dentin.[11]

Treatment of the dentition in dentinogenesis imperfecta includes the use of composite restorations, veneers, and full-coverage restorations.[12] Despite the advances in adhesive restorative techniques, the bond strength is compromised due to the malformed dentin structure and tubule architecture. However, adhesive restorations can be successful and remain a viable treatment modality. Dental intervention should take place as early as possible, with regular follow-up to identify and re-treat any failing restorations.

Attempts have been made to classify dentinogenesis imperfecta[13] into 3 types: type I, where dentinogenesis imperfecta is part of generalized disseminated OI (commonly OI types I, III, and IV); type II, where there is no association with OI; and type III (Brandywine isolate), which is isolated to a specific community in Maryland.

There have been reports of additional oral pathology in patients with OI; for example, a case report of a patient with OI who presented with multiple unilocular, bilateral, radiolucent lesions in the mandible.[14] These were diagnosed as multiple idiopathic bone cysts. There has also been a report of a large cementifying fibroma in a patient with OI[15] as well as familial associated gigantiform cementoma.[16]

It has been proposed that osseointegration be defined as a process whereby clinically asymptomatic rigid fixation of alloplastic materials is achieved and maintained in bone during functional loading.[17] This process depends on the state of the host bed and its capacity for healing. Initially, the implant must have good stability, which is dependent on the quality and quantity of the compact and cancellous bone. Subsequent osseointegration depends on the ability of the host to initiate controlled and coordinated contact osteogenesis at the bone-implant interface and distance osteogenesis at the surface of the osteotomy site.

Woven bone is formed before subsequent remodeling into lamellar bone. It might be surmised that in individuals with OI the quality and quantity of bone might affect initial stability due to its osteoporotic nature. In addition, the integration process might be impaired if the bone failed to remodel completely and remained immature woven bone.

Although there is very little in the literature relating to the use of dental implants in patients with OI, osteoporosis is well represented. A comprehensive review of literature relating to dental implants and implications of osteoporosis, as well as orthopedic interventions, has been published.[18] They conclude that subjects at risk for osteoporosis were not at increased risk of failure of implants to osseointegrate.

This observation is supported by case study evidence. For example, in one case report, a 5-year follow-up study of a female patient with severe osteoporosis rehabilitated with a 6-implant-supported fixed prosthesis was presented.[19] Animal studies, however, show that in experimentally induced osteoporosis, there are significant decreases in implant-bone contact and impaired extracellular matrix.[20] This finding may have an impact on long-term stability of implants, but its clinical significance is currently unclear.

The aim of this article is to provide insight into some of the issues that arose in the surgical and restorative rehabilitation of a patient with OI from the edentulous state. There is a paucity of reports in the dental literature on the use of implant-supported prostheses in such patients.

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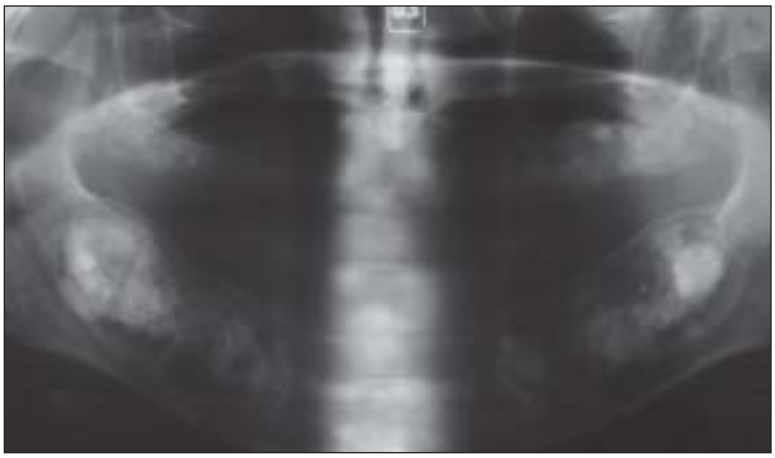

Fig 1 Preoperative dental panoramic radiograph.

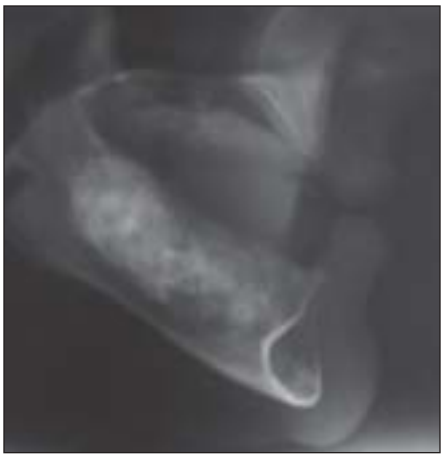

Fig 2 Preoperative lateral cephalometric radiograph.

CASE REPORT

A 34-year-old Caucasian woman with osteogenesis imperfecta (type IV) was referred to the implant clinic. She had been edentulous since the age of 14. She presented with failing restorations placed as a consequence of dentinogenesis imperfecta.

The medical history revealed several small bone fractures in childhood. The patient was normal in stature, with no significant associated deformity following healing of fractures. Her sclera were white.

This relatively young patient expressed a desire to replace her removable dentures with fixed implant restorations. The dentures the patient presented with were 11 years old and exhibited approximately 4 mm vertical occlusal wear, which was reflected in a slightly diminished vertical dimension of occlusion (VDO). The alveolar ridges were prominent, especially toward the posterior mandible. Even accounting for the occlusal wear, the interocclusal space was much smaller than would be expected for an individual who had been edentulous for 20 years. The dentures were necessarily thin with short-looking denture teeth, especially toward the posterior mandible.

Radiographic examination was carried out with dental panoramic and lateral cephalometric studies (Figs 1 and 2). The bone in the mandible had a thinner cortex and a less dense cancellous region than expected. The dental panoramic radiograph also showed diffuse radio-opaque masses related to the left and right body of the mandible. There was no radiographic expansion of either cortex in any direction. The inferior dental canal on either side was unaffected. These lesions were clinically asymptomatic. A diagnosis of a cemento-ossifying lesion, probably gigantiform cementoma, was made, and these were considered an incidental finding.

The replacement dentures provided incorporated a stable static occlusion based on centric relation, reestablishment of the lost VDO, and minor esthetic embellishments to meet the patient's esthetic requirements. The new dentures were copied to provide radiographic guides to enhance the information provided by computerized tomography (CT) scanning. CT scanning confirmed the need for sinus augmentation and veneer grafting of the anterior maxilla if a fixed implant prosthesis was to be considered. The CT scan also showed that there was insufficient interocclusal space between the posterior alveoli to accommodate fixed implant superstructures at the appropriate VDO. The following treatment plan was devised:

1. Bilateral reduction in mandibular height in the molar region prior to placement of implants to increase interocclusal space for the implant superstructure.
2. Reduction in height of the anterior mandible to compensate for its narrow profile and allow placement of implants in a suitable width of alveolus.
3. Bilateral maxillary sinus floor augmentations with veneer grafting to the anterior maxilla to improve lip support and increase bone volume for implant placement in positions determined by the recently constructed dentures.
4. Screw-retained maxillary and mandibular fixed implant-supported prostheses.

Maxillary grafting was carried out under general anesthesia. Corticocancellous bone blocks were harvested from the left iliac crest. Both maxillary sinus floor mucosal membranes were elevated without perforations via openings created in the lateral sinus walls. Blocks of autogenous bone were placed and

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Table 2 Implant Dimensions and Positions

Tooth position Universal (FDI)	Implant diameter (mm)	Implant length (mm)
4(15)	3.75	15
6(13)	3.75	15
8(11)	4.0	15
9(21)	3.75	15
11(23)	3.75	15
13(25)	3.75	15
20(35)	3.75	10
21(34)	3.75	11.5
24/25(31/41)	4.0	15
28(44)	3.75	15
29(45)	3.75	10

immobilized, and veneer onlay grafts were placed and immobilized to the anterior maxillary alveoli. All fixation was achieved using a 1-mm Wurzburg Titanium Microplate System and screws (Leibinger, Freiburg, Germany). Bio-Oss (Geistlich Biomaterials, Geistlich Pharma, Wolhusen, Switzerland) particulate allograft was placed in the voids around the grafts prior to closure.

At the same operation, the mandibular alveolus was reduced in height in the retromolar region. There was also reduction in height in the anterior part of the mandible to increase the crestal width to accommodate implants. There were no postoperative complications either in relation to the recipient site or donor site. Cefuroxime was administered pre- and postoperatively.

Implant placement was carried out 5 months later under general anesthesia with cefuroxime as a prophylactic antibiotic. Implant placement guides based on the radiographic guides (modified to fit the new alveolar contours) were used to inform implant position. Brånemark System Mk III Ti-Unite implants (Nobel Biocare, Göteborg, Sweden) were placed (Table 2). One plate was removed to allow for implant placement. It was noted that bone appeared macroscopically normal but was softer in quality. It was very easy to perform ridge dilatation at implant placement. Postoperative recovery was without incident.

Healing abutment connection was carried out 7 months later in the mandible and a further 2 months later in the maxilla. These procedures were carried out under local anesthesia. All implants were found to be clinically integrated. The postoperative period was without incident.

After all the surgical interventions, the fitting surfaces of the dentures were modified at chairside to accommodate changes in tissue contour and presence of healing abutments.

The restorations consisted of screw-retained gold frameworks, pink acrylic resin "alveolar replacement," and acrylic resin teeth. The restorative phase commenced 3 months after healing abutments were placed. Multi-unit abutments (Nobel Biocare, Göteborg, Sweden) were attached, with the exception of the midline mandibular region, where a 17-degree angled abutment was used. Abutments were tightened according to manufacturers' instructions to a torque of 35 Ncm.

In the maxilla there was perfect visible passive fit of the superstructure, but on final screw-tightening the patient could detect discomfort related to implants in the maxillary left premolar region. Two attempts were made to section, relocate, and solder the superstructure, but the discomfort on screw tightening persisted. There was no discomfort when the superstructure was sectioned in the midline and tightened in its 2 halves. It was therefore decided to complete the fabrication of the prosthesis in 2 parts. Mandibular procedures were uneventful.

The restorations were deemed satisfactory by clinician and patient, after some initial minor difficulty with "s" sounds was spontaneously overcome. Oral hygiene technique was prescribed using interdental brushes and thick floss.

The patient has had clinical and radiographic follow-up for 2 years. Function and esthetics remain entirely satisfactory. No pathologic peri-implant bone loss has been detected on periodic panoramic radiographs. Small deposits of supragingival calculus have been removed from the lingual aspects of the most anterior 3 transmucosal abutments using hand instruments at regular maintenance appointments at intervals of 4 months. The peri-implant soft tissue condition has remained healthy in appearance, with no deepening of peri-implant probing depths. It is expected that the acrylic resin components of the superstructures will require refurbishment at some time over the next 5 to 10 years (Figs 3 to 6).

DISCUSSION

A search of the English-language literature highlighted 3 cases that differ significantly from that presented here. There is a reported case of DI involving a staged sinus augmentation and multiple implant placements to support fixed prostheses in all quadrants of the mouth.[21] The patient remained partially dentate, with subsequent implant placements as fur-

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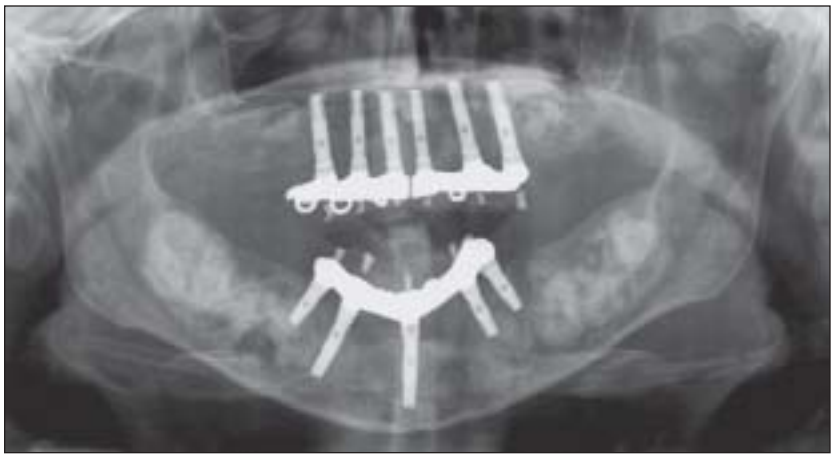

Fig 3 Postoperative dental panoramic radiograph.

Fig 4 Postoperative facial photograph with teeth in occlusion.

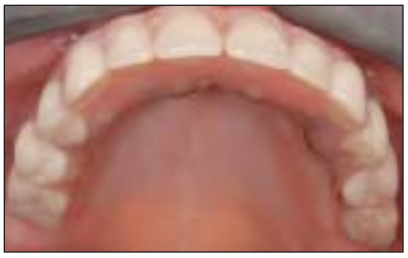

Fig 5 Postoperative occlusal photograph of maxilla.

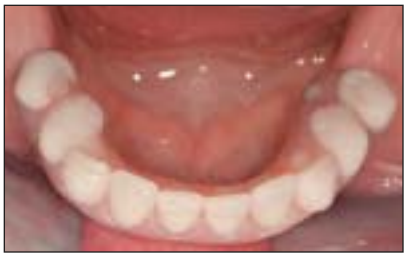

Fig 6 Postoperative occlusal photograph of mandible.

ther teeth were lost due to the effects of DI. Bone augmentation was carried out by harvesting iliac crest bone via a large-bore needle in a closed procedure. This was further augmented with bone powder (Dembone) and artificial bone (Osteograf). Two implants in the maxillary molar region failed and were removed at 3 years. Mobility of implants in the maxillary canine and premolar regions was also reported, but apparently the prostheses remained stable. The authors also reported the fracture of an implant in the maxillary premolar region; the implant was removed and replaced.

The second case was a case of bone augmentation and dental implant rehabilitation confined to a single mandibular quadrant.[22] The patient was affected by classic type I OI with associated DI. The mandibular right quadrant was augmented in a buccolingual direction with autogenous bone harvested from the right mandibular ascending ramus and used as an onlay graft. The authors described the postoperative period as uneventful. The patient was then restored with a 2-implant-supported 3-unit fixed partial prosthesis.

There is also a case report of oral rehabilitation with implant-supported removable overdentures in the maxilla.[23] This was carried out following modified Le Fort I osteotomy and osteodistraction to correct dysgnathia due to a hypoplastic maxilla. The prosthesis, which was supported by 5 implants, was placed subsequent to bilateral sinus floor elevation and alveolar process augmentation with autogenous iliac crest bone.

The present case differs significantly in being full-mouth rehabilitation from the edentulous state. The patient had bilateral cemento-ossifying lesions, which were an incidental finding. Although unlikely to have contributed to the need for reduction in the bulk of her posterior mandible, they may have had an impact on the quality of bone in which implants were placed.

The patient was restored with a shortened dental arch. This was directly related to the space limitations, which limited the sites available for implant placement. Surgical guides, fabricated from optimized complete dentures, facilitated prosthetically driven implant placement and thus the restorative phase.

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The height of the acrylic resin needed for the mandibular prosthesis led to a slight compromise in esthetics. This increase in acrylic resin was needed following reduction in alveolar height to allow implant placement in bone of appropriate width as an alternative to further bone grafting to widen the alveolus.

In conclusion, this case shows implant-supported fixed prostheses to be a viable treatment option for an edentulous patient with osteogenesis imperfecta, albeit with relatively short follow-up to date. Surgery involved augmentation of bone in the maxillary sinus floor and maxillary anterior regions as well as reduction in bone bulk in the mandible to accommodate the restorative superstructure.

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