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# Dentinogenesis Imperfecta: Full-Mouth Rehabilitation using Implants and Sinus Grafts – A Case Report

**Abstract:** This case report outlines one possible treatment modality to manage the developmental abnormality dentinogenesis imperfecta (DI). In this case, the patient's dentition is restored using a combination of full-coverage crowns for the remaining teeth and implant-supported crowns to replace missing teeth in a re-organized occlusal scheme. The case also demonstrates the effective use of the sinus graft procedure with simultaneous placement of dental implants. This paper also aims to make the reader aware of the current thinking behind treatment delivered to this group of patients, focusing on full-mouth rehabilitation using a combination of implant-supported and conventional metal ceramic crowns.

**Clinical Relevance:** For the general dental practitioner this case outlines the prevalence and cause of DI. It demonstrates how early diagnosis and appropriate referral has an impact on future treatment.

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Dentinogenesis imperfecta (DI) is a genetically determined developmental defect of dentine. The condition is broadly grouped into three categories:<sup>1</sup>

1. Type I: the dental manifestation of osteogenesis imperfecta.
2. Type II: classical hereditary opalescent dentine.
3. Type III: Bradywine isolate opalescent dentine.<sup>2</sup>

The prevalence of DI is 1:8000.

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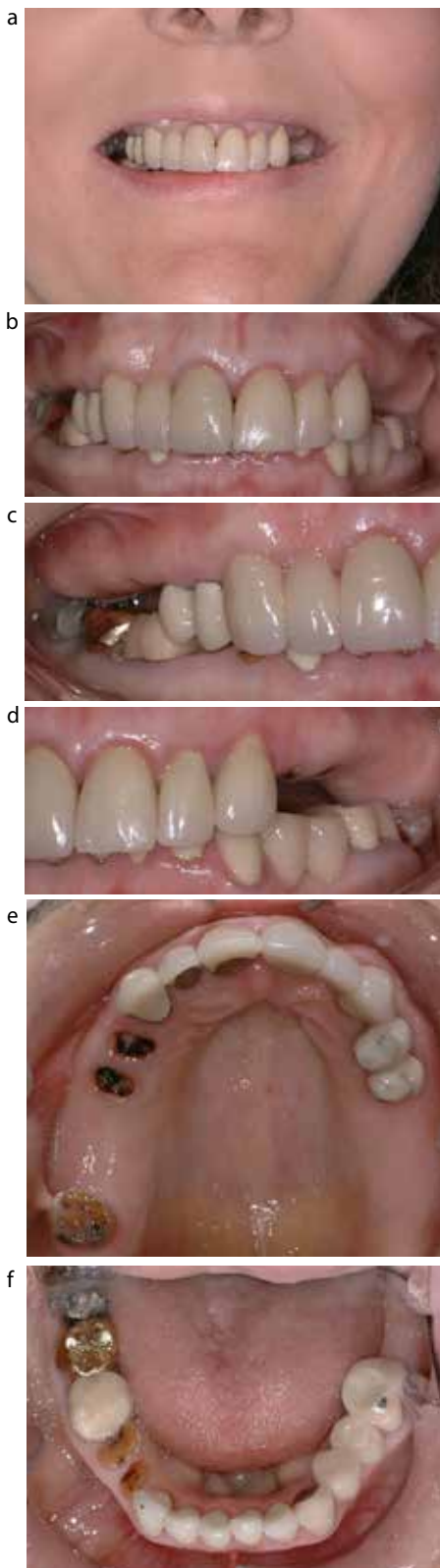
Type I occurs as part of osteogenesis imperfecta, which is caused by mutations in collagen (Type 1 collagen alpha 1 and alpha 2 chains) genes. The inheritance pattern of DI type II and type III is autosomal dominant. Mutations in the DSPP gene have been identified in both types of DI occurring as an isolated trait. Research indicates that Type II and Type III are different expressions of the same gene.<sup>3</sup>

Histologically, the dentine appears normal. However, the scalloping at the dentino-enamel junction is missing. This scalloping acts to hold the two tooth structures together mechanically; in its absence enamel is easily chipped off the dentine.<sup>4,5</sup> Clinically, teeth appear bluish-brown and opalescent. The enamel tends to chip and wear away, exposing the malformed abnormal dentine which wears rapidly. Radiographically, the crowns are bulbous while the roots are short and thin. The pulp chambers are wide initially but obliterate soon after eruption.

## Treatment in the primary dentition

The aims of treatment are to remove sources of infection or pain, restore aesthetics and protect posterior teeth from wear.<sup>6,7</sup> Patients present with this condition affecting their teeth to various degrees so it is not possible to make generalized treatment plans. In general, all patients require a thorough preventive regime, including diet advice and oral hygiene instruction.

Restoration of the dentition ranges from the simple placement of resin-modified glass ionomer cement in areas of enamel loss in the very young and unco-operative child, to placement of stainless steel crowns and direct composite for the more co-operative child.<sup>8</sup> Placement of these crowns is advised where there is more extensive tissue loss. Where tissue loss is more severe and little is remaining above the gingivae, extraction of the primary tooth is indicated. If the teeth become non-vital,



**Figure 1.** (a) Extra-oral view pre-treatment; (b) intra-oral view pre-treatment; (c) right lateral view; (d) left lateral view; (e) upper arch; (f) lower arch.

or abscess formation occurs, primary tooth pulp treatment is contra-indicated and again extraction is necessary.<sup>9</sup>

### Mixed dentition

As the permanent dentition develops it should be monitored closely for tooth surface loss. The amount and distribution of the loss will dictate the type of treatment at this stage. The majority of patients may be treated with cast occlusal onlays on the molar and premolar teeth as this minimizes tooth surface loss whilst maintaining the correct OVD.<sup>10</sup> This is also a minimally invasive technique which is of paramount importance at this stage of the patient's dental development.

Aesthetics is normally a concern as the permanent incisors erupt. In the short term, the treatment of choice is direct composite veneers. These are minimally invasive, aesthetic and also help towards better oral hygiene as the irregular chipped enamel surface is now replaced by a polished composite surface.

### Permanent dentition

Again, the amount and distribution of tooth surface loss will dictate the level of treatment. At this stage, many patients are considered for full-mouth rehabilitation. However, there are some major differences when treating this group of patients compared to the general population. Classically, these patients have teeth with short, thin roots which have a decreased root surface area available to distribute physiologic forces generated during function.<sup>11</sup> However, this is not a contra-indication to crowning the teeth.

Endodontically, the teeth have obliterated pulp chambers and canals. Loss of vitality and subsequent development of apical pathologies are rare,<sup>12</sup> however, if orthograde endodontic treatment is indicated it is often technically impossible, generally resulting in tooth loss. Retrograde root treatment is another option but should be contra-indicated for teeth with short roots.<sup>13</sup>

### Case report

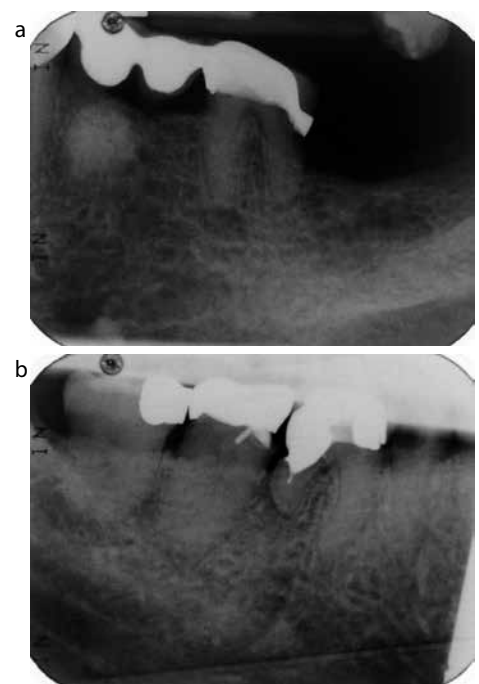
The 48-year-old female patient was referred by her GDP complaining of

an unstable and unretentive upper partial denture. She gave a history of brown, mottled teeth since childhood that were very brittle and wore easily. She had a long history of dental treatment ranging from simple fillings to complex crown and bridge work (Figure 1). Her main wish was to have fixed replacement of her missing teeth rather than a removable partial denture.

Medically, the patient had controlled hypertension. She was also a non-smoker. She had no apparent family history of the condition affecting her teeth.

Clinically, the patient had a heavily restored dentition. Bridges were present UR3–1 replacing the UR2 and LL3–6 replacing the LL4 and 5. Crowns were present on the UR5,4, UL1,2,3, LL1,2, LR1,2,3,6,7. Retained roots were present UL4,5 and retained LRD,E were present whilst the LR4,5 were developmentally absent.

The patient had a Class II division 1 incisal relationship with a 7 mm overjet and increased overbite. Owing to the loss of tooth structure over the years, the patient was apparently overclosed. In the maxilla an acrylic denture replaced the missing teeth. The denture was tissue-supported. Clinically it was unretentive and unstable, and the patient found it difficult to tolerate. The patient's oral



**Figure 2.** (a) LL6 pre-treatment; (b) lower right quadrant pre-treatment.



**Figure 3.** Lateral approach for sinus graft. Note, different patient from case report.

hygiene was good with a BPE score of 0 in all but the lower anterior sextant, which scored 2 due to supra-gingival calculus deposits.

Radiographic examination revealed general root canal obliteration of the remaining teeth and root morphology characteristic of dentinogenesis imperfecta (Figure 2). It also showed caries in the LL6 bridge abutment, a mesial marginal gap in the crown restored LR7, retained roots UL4,5 and retained LRD,E. No periapical pathology was present and bone levels were normal.

The diagnoses reached were as follows:

- Dentinogenesis imperfecta Type 2;
- Retained roots UL4,5;
- Retained deciduous teeth LRD, E/ developmental absence LR4, 5;
- Caries and failing bridge LL3–6;
- Failing crown LR7;
- Failing P/-;
- Reduced lower face height;
- Missing permanent teeth UR8,7,6, UL6,7, LL7,8.

The initial treatment plan was to restore caries and stabilize her oral health. Treatment options discussed with the patient for restoration of the missing teeth included the fabrication of new upper and lower partial dentures, fixed conventional bridges or implant-supported crowns. The patient was keen to have a fixed solution and opted for implants at an early stage. The initial treatment plan and interpretation was as follows:

- Oral hygiene instruction;
- Scale and polish;
- Extraction UL4,5 and LRD, E;
- Removal failing fixed prosthesis to assess if restorable and temporization LR7, LL3, LL6;
- Articulated study models in retruded arc of closure, using a facebow record. Diagnostic

wax-up at a reorganized occlusal scheme giving full interdigitation, incisal guidance and canine guidance in lateral excursions and an increased occlusal vertical dimension;

- Radiographic stent UR6, UL4,5,6;
- Cross-sectional radiographs UR6, UL4,5,6 and LRD,E areas.

After the initial treatment and interpretation were completed it became apparent that the LL6 was unrestorable, so the bridge in the area would need sectioning, leaving the LL3 as a crown. It also became apparent that the UL3 was unrestorable due to insufficient coronal structure and would require removal. The wax-up was discussed with the patient and the definitive treatment plan was completed. It was as follows:

- Extraction LL3 and LL6;
- Implant placement UL3,4 (Friadent Xive® implants);
- Construct upper and lower temporary partial dentures;
- Cross-sectional radiographs with radiographic stents *in situ*;
- Right and left maxillary sinus grafts and simultaneous implant placement under local anaesthetic UR7,6 and UL5,6;
- Implant placement LR4,5,6 and the LL4,5,6;
- Reorganizing the occlusion by increasing the occlusal vertical dimension by 2 mm in the retruded position of the mandible;
- Restore implants with screw retained restorations using new occlusal scheme and the diagnostic wax-up as a template;
- Restoration of remaining dentition at the reorganized occlusion.

The treatment plan was carried out over the course of a year.

## Single vs 2-stage sinus graft procedures

The posterior maxilla is often a difficult area to restore using implants as there may be insufficient bone height between the floor of the maxillary sinus and the oral cavity, rendering traditional placement of implants impossible. Sinus grafting is a procedure which allows the floor of the sinus to be elevated and bone grafted in to the site to facilitate placement of dental implants. This is recognized as a predictable and viable treatment modality.<sup>14</sup>

Two-stage sinus lifting involves first lifting the sinus floor with either autogenous bone or xenograft bone substitutes (eg Bio-Oss®), after which three

months of healing is allowed, followed by a further procedure to place dental implants. In some cases, it is possible to combine the sinus lift procedure with implant placement in a combined procedure. This is indicated when there is at least 4 mm of bone available under the sinus floor to give initial primary stability for implant placement. It is critical that primary stability of the implants is achieved. If residual bone height is less than 4 mm and/or primary stability of the implant is inadequate, a 2-stage sinus graft procedure is necessary.<sup>15</sup> The advantages of the single stage procedure are decreased treatment time and reduced morbidity due to the decreased number of surgical stages undertaken.

The autogenous bone graft has long been considered the gold standard.<sup>16</sup> The bone is either harvested from an intra-oral or extra-oral site. Intra-oral sites include the maxillary tuberosity, the ramus and mental region of the mandible. Extra-oral sites include the iliac crest, tibia and calvarium. One disadvantage of these sources of bone is the morbidity from the donor site.

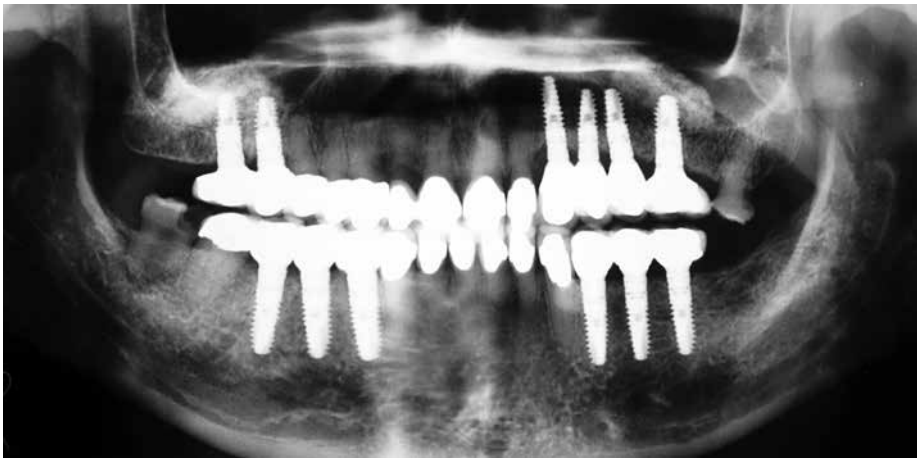
It is now commonplace to use bovine bone alone to graft the sinus or a composite autogenous bone-xenograft bovine bone graft. In this regard, it has been shown that bovine bone alone is a successful material for sinus graft procedures.<sup>17,18</sup>

The technique used in this case was a single stage procedure. Flap design was a three-sided, full thickness mucogingival flap giving access for a lateral approach (Figure 3). The graft used was locally harvested bone chips and Bio-Oss® with Bio-Gide® being placed over the access window in the sinus.<sup>19</sup> Good primary stability of the implants was gained and the post-operative radiographs showed good bony infill around the implants placed in the sinus graft (Figure 4).

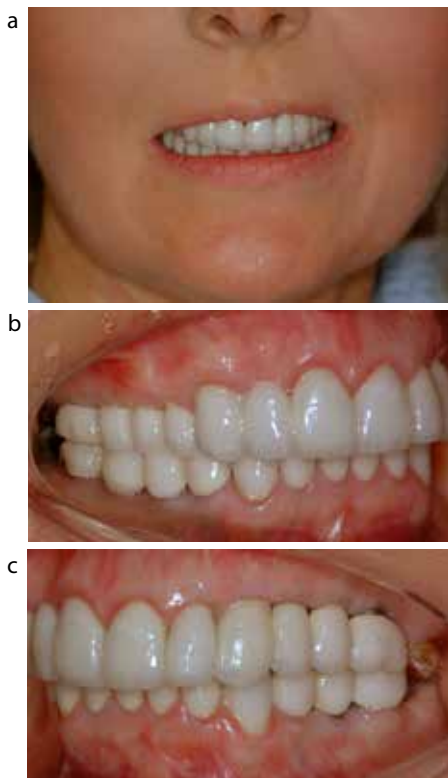
## Overall treatment

The treatment was split into manageable segments. The first major aspect was removal of unrestorable teeth and construction of some partial dentures. This was followed by staged surgical and review appointments in which implants were placed and the bilateral sinus grafts were undertaken (Figure 5).

This was then followed by removal of the old metal ceramic crowns on the remaining teeth, preparation and then temporization. The teeth were temporized



**Figure 4.** Post-treatment implant crowns. Note patient had lost LR8 filling on review.



**Figure 5.** (a) Post-treatment extra-oral view; (b) right lateral view; (c) left lateral view.

with lab-made composite prototype crowns at the new occlusal scheme as they would be *in situ* a number of months to allow for restoration of the implants and minor adjustments to the occlusal scheme.

The next stage was to expose the implants and placement of the temporary crowns at the new occlusal scheme, again using the diagnostic wax-up as a guide to occlusal dimensions. This was completed over

a number of visits, with the occlusion being adjusted as necessary. Once the implants were temporarily restored to the reorganized occlusion, the final metal ceramic crowns of the natural teeth were cemented. These then acted to stabilize the new occlusion while restoration of the implants was completed.

The patient was happy with the result and has adapted to her new occlusal scheme very well. On review, the patient had no major concerns. Her long-term dental care will be carried out by her original GDP and her implants will be reviewed annually. It is worth mentioning that the treatment was completed in April 2009, therefore it is still too early to call the case a long-term success.

## Discussion

One of the most important aspects in the treatment of DI is early recognition and diagnosis of the disease and referral to the appropriate specialty. The type of treatment indicated is largely dependent on the age of the patient when first seen,<sup>20</sup> but generally involves thorough prevention and protection of the relatively weak tooth structure.

Although no formal study exists comparing the complexity of treatment to the age of initial presentation, it is commonly understood that early management and strict long-term follow-up can lead to a less invasive approach than has been shown in this case. It has been illustrated that a delay in implementing the correct treatment can lead to future treatment being more complex.<sup>21</sup>

As shown, the complex nature

of treatment outlined was not only dictated by the genetic condition, but also by the comparatively late presentation of the patient. At the time of presentation, the patient already had numerous missing dental units and multiple teeth restored with crowns/bridges. This immediately removes the possibility of minimally invasive procedures, and commits the patient to fixed/removable prosthesis. In this case, the patient was not keen to continue down the removable prosthesis pathway and so implant restoration of her missing units was used.<sup>22,23</sup>

It should be noted that, in this case, the patient suffered from DI Type II, rather than DI Type I that is associated with osteogenesis imperfecta. In Type II, DI is the only trait and bone quantity and quality is the same as an unaffected individual. Although some anecdotal evidence exists regarding implant placement in patients with osteogenesis imperfecta, no long-term controlled studies have been carried out.

This case demonstrates that oral rehabilitation of a patient suffering from Type II dentinogenesis imperfecta using tooth-supported and implant-supported restorations is a viable and clinically effective option.

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## Book Review

**Contemporary Esthetic Dentistry.** By George Freedman and contributors. London: Elsevier Health Sciences, 2012. ISBN 978-0-3230-6895-6.

This nearly 800 page book is a terrific collection of comprehensively referenced work by Dr George Freedman who has taken contributions from over 50 very experienced names in the world of Esthetic Dentistry.

It is beautifully illustrated and this quality is consistent throughout pretty much all of the book.

What I especially liked is that virtually every aspect of clinical dentistry has been touched on with an aesthetic slant or view. Each chapter, although written by many different names, still possesses a thread of the headlines that run through the book, namely: Relevance to Esthetic Dentistry, Clinical Considerations, Material Options,

Artistic Elements, Evidence Base, Controversies and Future Developments. This thought process has been applied to 34 different chapters ranging from: Caries Management, Adhesion, Anterior and Posterior Composites, Photography, Bleaching, Direct Veneers, Porcelain Veneers, Denture Esthetics, even to Sterilization and Disinfection.

Dr Freedman candidly admits that dentistry moves on rapidly in the time it takes to put the book together and new developments can change treatment options and trends quickly, but this is also a 'live' book which is constantly being added to online.

I felt that Orthodontics could have been represented in more detail, even if the book was put together 2–3 years ago. And some elements on the psychology behind smile design and patients' expectations in different parts of the world might have been quite

interesting too. I would also like to see something on Progressive Smile Design. But I am nit-picking here because this is a vast piece of work and Dr Freedman should be commended for putting together something as comprehensive as this.

Stand out chapters for me were William Mopper and Sunil Bhoolabhai on Anterior Composites and one of Dr Freedman's own chapters on Ultraconservative Dentistry, which I thought was excellent.

Don't expect to plough through this book on a trip or holiday. It is a reference book and will be particularly useful sitting in your practice and much of it will be relevant for many years to come. I consider it a good buy for any dentist interested in carrying out his/her dentistry aesthetically and we all know that ought to apply to every dentist.

**Dr Tif Qureshi, President, The British Academy of Cosmetic Dentistry**