

Multidisciplinary approach for a patient with dentinogenesis imperfecta and anterior trauma

Won-Jong Roh,^a Seung-Goo Kang,^b and Su-Jung Kim^b

Seoul, Korea

Dentinogenesis imperfecta is an inherited dentinal dysplasia involving several risks for orthodontic treatment. This case report describes the multidisciplinary treatment of a 17-year-old girl whose Class II Division 1 malocclusion was complicated by dentinogenesis imperfecta type II and maxillary anterior trauma. (Am J Orthod Dentofacial Orthop 2010;138:352-60)

Dentinogenesis imperfecta (DI) is an inherited disorder of tooth development. Affected teeth are discolored and weaker than normal. The disorder is divided into 2 types. Type I is associated with a collagen defect,^{1,2} and type II is caused by a disorder of dentin mineralization. Therefore, type II is not associated with generalized conditions such as osteogenesis imperfecta (OI). For the successful treatment of patients with DI, not only is a differential diagnosis between types I and II necessary, but also a multidisciplinary approach should be considered. This case report illustrates the multidisciplinary treatment of a patient with type II DI, delineating some therapeutic guidelines for orthodontic management.

DIAGNOSIS AND ETIOLOGY

A 17-year-old girl was referred to the department of orthodontics in KyungHee University Medical Center after she had received emergency treatment of resin-wire splinting on her maxillary incisors due to the trauma. Her chief complaints were unsightly discoloration of the teeth and maxillary incisor protrusion. Her family history supported the provisional diagnosis of DI, with a pedigree that spanned 2 generations on her maternal side. Her mother had undergone full-mouth rehabilitation because of DI, and therefore wanted her daughter to have prosthetic reconstruction as her final treatment. The patient was in good physical

condition. Her medical history showed no evidence of OI, such as a history of frequent fractures of long bones, laxity of joints, blue sclera, hearing loss, or increased bleeding tendency.^{3,4}

Pretreatment photographs (Fig 1) showed a dentition with brown to gray opalescent teeth without severe attrition. She had a Class II Division 1 malocclusion with crowding, anterior open bite, large overjet, palatally blocked maxillary left second premolar, and multiple caries. Dental casts confirmed asymmetric right and left occlusal curves caused by the extrusion of maxillary right first molar (Fig 2). A panoramic radiograph showed impaction of the mandibular left second molar and the 4 third molars in addition to the features of DI: short roots, bulbous crowns, cervical constrictions, and pulpal obliteration (Fig 3, A).¹⁻⁴ In the periapical film, tooth fractures on both maxillary central incisors were confirmed without alveolar bone fracture (Fig 3, B). A lateral cephalometric radiograph and its tracing showed a skeletal Class II hyperdivergent pattern (Fig 4).

TREATMENT OBJECTIVES AND PLAN

Integrated conservative, orthodontic, periodontal, and prosthodontic treatment objectives were established as shown in Table I. The ultimate objective was to achieve an esthetic reconstruction to conceal the discoloration. Full-coverage restorations were planned except for the posterior teeth, because this patient had neither severe attrition nor reduced vertical dimension.

The specific objectives of orthodontic treatment included retrusion of the maxillary anterior teeth and upper lip, maintenance of the sagittal position of the mandibular incisors, alignment of the maxillary and mandibular teeth, vertical control of the asymmetric occlusal curve, and establishment of a Class II molar relationship with proper overjet and overbite. The orthodontic treatment plan involved extraction of the

From the Department of Orthodontics, School of Dentistry, KyungHee University, Seoul, Korea.

^aClinical director.

^bAssistant professor.

The authors report no commercial, proprietary, or financial interest in the products or companies described in this article.

Reprint requests to: Su-Jung Kim, Department of Orthodontics, School of Dentistry, KyungHee University, 1 Hoegi-Dong, Dongdaemoon-Ku, Seoul 130-701, Korea; e-mail, ksj113@khu.ac.kr.

Submitted, July 2008; revised and accepted, December 2008.

0889-5406/\$36.00

Copyright © 2010 by the American Association of Orthodontists.

doi:10.1016/j.jado.2008.12.023



Fig 1. Pretreatment facial and intraoral photographs.

fractured maxillary central incisors, the maxillary left second premolar, the hopeless mandibular right first molar, the impacted mandibular left second molar, and the 4 third molars. To preserve the alveolar ridge, extractions of the maxillary left central incisor and the mandibular right first molar were postponed until they were replaced by prostheses. The maxillary right lateral incisor, canine, and first premolar would substitute for the maxillary right central incisor, lateral incisor, and canine, respectively.

TREATMENT ALTERNATIVES

Our treatment plan involved atypical extractions. In the maxillary arch, another option would have been to extract the right and left first premolars to reduce the anterior protrusion and overjet. However, on the right side, extraction of the first premolar would decrease the number of remaining teeth and require additional

implant surgery for the lost central incisor. Orthodontic traction of the palatally blocked left second premolar into the extracted first premolar site would have a periodontal advantage, but we needed reinforced anchorage on the left side so as not to deviate the upper midline to the right during the mesial movement of right lateral incisor, canine, and first premolar.

In the mandibular arch, surgical exposure and orthodontic traction of the impacted left second molar seemed to be reckless, because of its poor direction and position. Implants or prosthetic reconstructions for lost second molars would have been unnecessary considering the Class II molar relationship as the final occlusion.

TREATMENT PROGRESS

Before the orthodontic treatment, the patient and her mother signed an informed consent document that

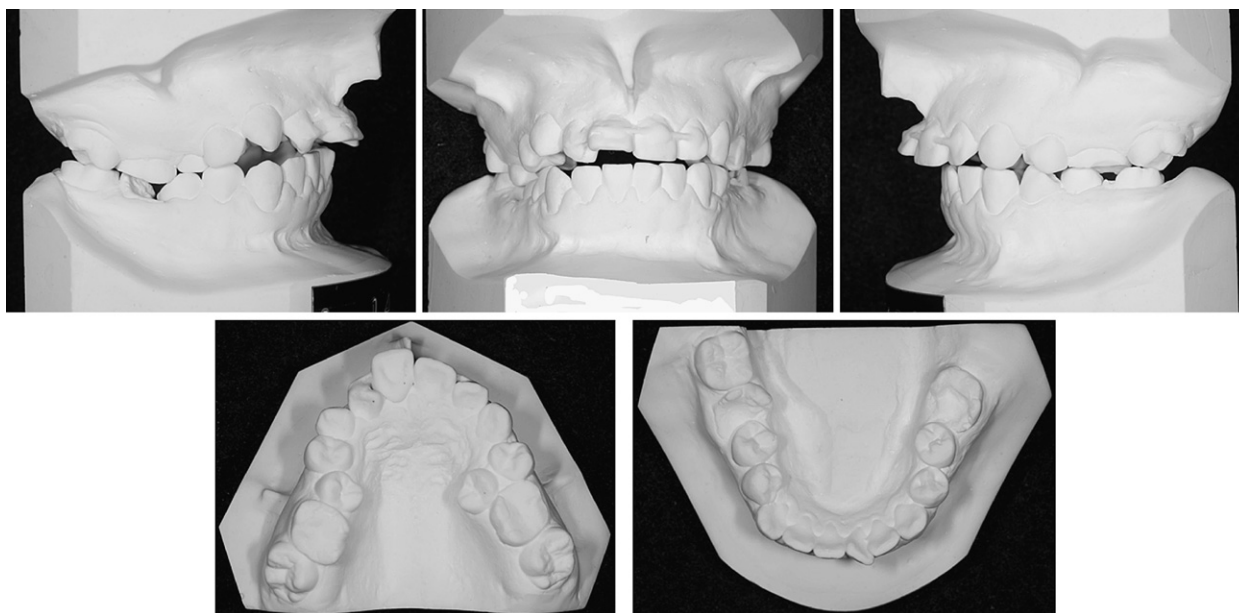


Fig 2. Pretreatment dental casts.

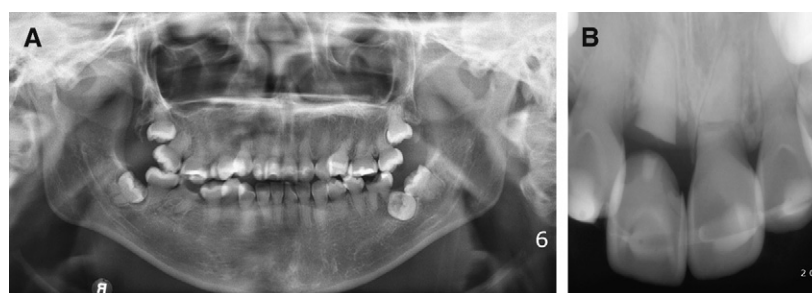


Fig 3. **A**, Panoramic radiograph and **B**, periapical radiograph of the maxillary incisors at pretreatment.

explained the treatment risks such as root resorption, sheering off or loss of enamel, tooth fractures, and increased tooth discoloration.

After restoration of the carious teeth and extraction of the maxillary right central incisor and left second premolar, conventional direct bonding of brackets (MBT, 3M Unitek, Monrovia, Calif) was performed on all teeth except the molars (Fig 5). The maxillary right lateral incisor, canine, and first premolar were moved mesially and sequentially by using a miniscrew (Orlus, Osolution, Seoul, Korea) on the maxillary left side. Vertical leveling of the asymmetric occlusal curve with extrusion of the maxillary anterior teeth was obtained by using a second miniscrew in the right mandibular region (Fig 6).

On the panoramic radiograph taken after leveling (Fig 7), markedly increased root resorption of the

mandibular right second molar was observed, associated with progressive eruption of the remaining third molars. Therefore, the original treatment plan was altered to include extraction of the mandibular right second molar.

After debonding all brackets (Figs 8 and 9), the patient was referred first to the endodontist for follow-up of the traumatized maxillary incisors and then to the periodontist for crown lengthening of the maxillary right lateral incisor and a labial frenectomy. In addition, ridge augmentation by free gingival graft was performed on the depressed residual ridge in the maxillary left central incisor area (Fig 10). Finally, all ceramic crowns and bridges were constructed, and an implant was placed in the mandibular right first molar area. No orthodontic retainer was used.

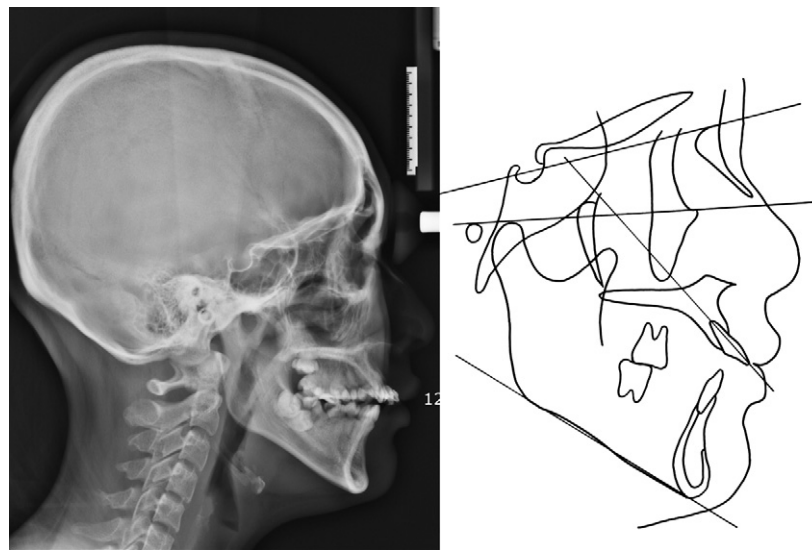


Fig 4. Pretreatment lateral cephalogram and its tracing.

Table I. Integrated treatment objectives and plan

Conservative objectives	Restore carious teeth Follow up the traumatized teeth
Orthodontic objectives	Improve facial and dental esthetics Retrude maxillary anterior teeth and upper lip Maintain sagittal position of the mandibular incisors Decrowd the dentition Level the asymmetric occlusal curve Establish Class I canine and Class II molar relationships Obtain functional occlusion by proper overjet and overbite
Periodontal objectives	Detach the heavy labial frenum Lengthen the crown of the substituted tooth Replace the hopeless mandibular first molar with an implant
Prosthetic objectives	Improve the dental esthetics; reconstruct the discolored teeth Establish the final occlusion Maintain the vertical dimension

TREATMENT RESULTS

The postorthodontic treatment photographs (Figs 8 and 9) show good interdigitation of Class II molar relationships, reduced overjet, and improved overbite. The remaining space on the distal surface of the maxillary left lateral incisor would be corrected by the prosthesis. Symmetric flattening of the occlusal plane was accomplished. Arch symmetry was established despite the asymmetric extractions and treatment mechanics. The posttreatment photographs (Fig 11) show a pleasing facial appearance with retraction of

the upper lip. A more esthetic level of gingival lines of the maxillary incisors was achieved. The corrected midline at debonding was shifted slightly to the right side after prosthetic reconstruction; however, the esthetic improvement left the patient highly satisfied with her full smile. The posttreatment cephalogram and its tracing (Fig 12) showed retraction of the maxillary incisors, with the sagittal positions of the mandibular incisors maintained. Root resorption of the maxillary left lateral incisor was noted in the posttreatment panoramic radiograph (Fig 13). Overbite was increased by the extrusion of both maxillary and mandibular anterior teeth, preserving the vertical dimension (Table II).

DISCUSSION

This case highlights some important issues related to DI that are relevant for all orthodontists. First, the clinician must differentiate between the 2 types of DI because type I, which is accompanied by OI, requires more comprehensive management. Shields et al¹ classified DI into types I, II, and III. Although the dentin in types I and II is clinically, radiographically, and histologically similar, patients with type I DI can also suffer from generalized conditions such as osteoporosis, bowing or fractures of the long bones, joint laxity, increased bleeding tendency, and allergic response to latex.⁵⁻⁷ All of these conditions significantly impact orthodontic treatment. Clinicians can also distinguish type I by its distinctive craniofacial traits: triangular-shaped face, broad bossed forehead, basilar invagination, short neck, midface hypoplasia, underdeveloped maxilla, and large tongue. Additional characteristics of type I



Fig 5. Intraoral photographs after passive bypassing archwire (0.016 × 0.022-in stainless steel) was placed. The right central incisor was extracted, and the space was closed by mesial movement of the right lateral incisor with miniscrew anchorage.



Fig 6. Intraoral photographs during the asymmetric leveling and anterior extrusion: an 0.018-in stainless steel archwire with T-loop preactivated by a gable bend was placed, and vertical elastics were used from the T-loop to the mandibular miniscrew.

DI that affect treatment include a high incidence of Class III malocclusion, anterior and posterior crossbite, posterior open bite, and molar impaction.^{4,8} Our patient was diagnosed with type II DI because she had no physical, facial, or radiographic signs associated with OI.

Second, a multidisciplinary approach to the patient from diagnosis to treatment was the key to the favorable outcome for this patient, because of the complications of severe orthodontic problems and many teeth destroyed by caries and trauma. Most patients with DI expect the discoloration, severe attrition, and enamel fractures of their teeth to be ultimately corrected by full-mouth rehabilitation. Although the overall risk of caries and periodontal disease appears to be normal in a patient with DI, for more complex esthetic and functional reconstruction, preprosthetic collaboration among conservative orthodontic, periodontal, and surgical plans is especially important.⁸

Third, bonding the brackets was successful in our patient; this was achieved without bonding failure during treatment or enamel loss after rebonding or debonding. It has been proposed to use bands on all teeth

cemented with glass ionomer cement, rather than using bonded brackets, to reduce the risk of enamel fracture.^{9,10} However, it has also been reported that adhesive dentistry is indicated for patients with DI, and successful bonding of orthodontic brackets has been accomplished.^{4,11} Despite these positive reports, it was not determined whether enamel fractures occurred later during the orthodontic treatment period. In our patient, before the clinical application of direct bonding, we performed a preliminary test using her extracted maxillary right central incisor. Both the preliminary test and the patient outcome showed that the bond strength of brackets with Transbond XT (3M Unitek) was as sufficient as for normal orthodontic patients, and that there was no enamel damage after conventional debonding.

Fourth, orthodontic miniscrew anchorage in patients with DI is sufficiently stable to be used throughout treatment. Miniscrews were used in our patient for efficient asymmetric space closure and control of the asymmetric occlusal curve. Based on a study that bone healing is essentially normal in the normal time frame, in spite of poor density similar to the original

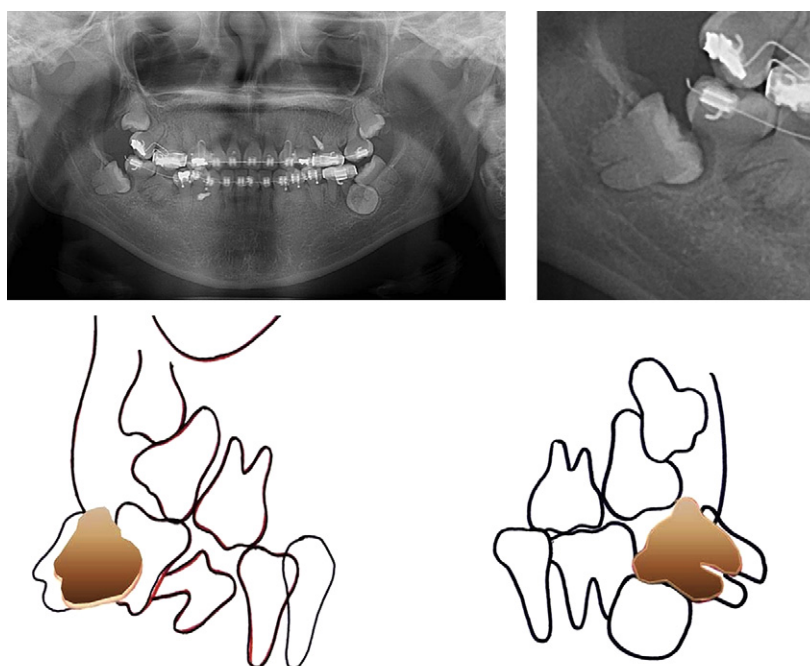


Fig 7. Panoramic radiograph after leveling, with enlarged section to show the progressive root resorption on the mandibular right second molar. Superimposition with pretreatment panoramic radiograph shows progressive eruption of the remaining third molars.



Fig 8. Intraoral photographs after debonding.

quality,¹² various procedures such as orthopedic appliances,⁴ orthognathic surgery,^{3,13,14} and dental implant surgery^{15,16} have been used even for patients with type II DI. Prabhu et al¹⁶ reported that dental implants were successfully osseointegrated in a patient with type I DI who was prone to mandibular fractures. Therefore, orthodontists can safely use miniscrew anchorage in patients with both types of DI.

Finally, the risk of root resorption proved to be high in this patient. Although there was no generalized apical root resorption, lateral root resorption associated with a developing adjacent tooth was remarkable.

Superimposition of panoramic radiographs (Fig 7) showed considerable eruption of the remaining third molar, resulting in external root resorption with little movement of the mandibular second molar. This outcome is unusual in conventional orthodontic patients. Therefore, when treating patients with DI, earlier extraction of the developing third molars should be considered. Although the rate of tooth movement was normal in our patient, and the short conical roots showed no signs of abnormal tipping or mobility, we recommend keeping the active treatment period as short as possible because of the risk of root resorption.

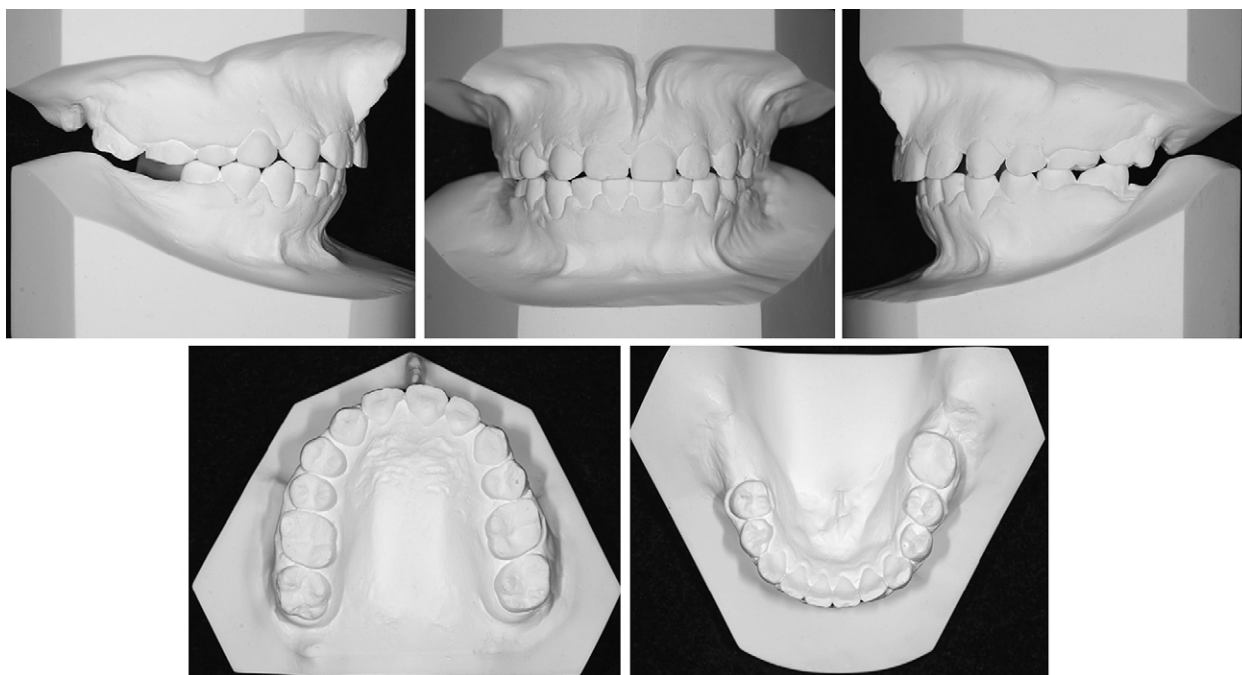


Fig 9. Dental casts after debonding.

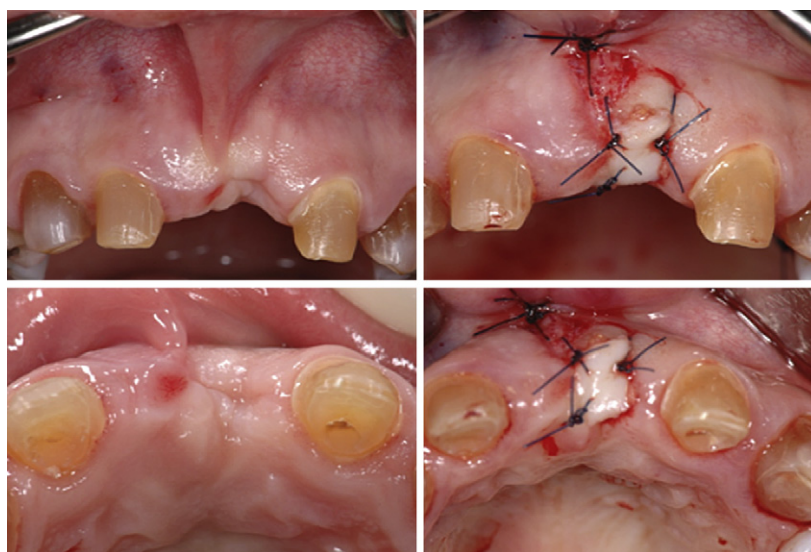


Fig 10. Intraoral photographs of the maxillary anterior region. Ridge augmentation by free gingival graft and frenectomy of heavy labial frenum were performed.

CONCLUSIONS

This report focused on the importance of a multidisciplinary approach and specific orthodontic considerations for the successful treatment of a patient with DI. Adhesive dentistry, periodontal surgery, implant-supported prostheses, orthodontic treatment, and orthognathic surgery

might be indicated in these patients. In the orthodontic aspects, the conventional procedure of bonding brackets and miniscrew anchorage is available for patients with DI. Proper management should be guided by differential diagnosis between the types of DI, leading to improved function and esthetics.



Fig 11. Facial and intraoral photographs after prosthetic reconstruction.

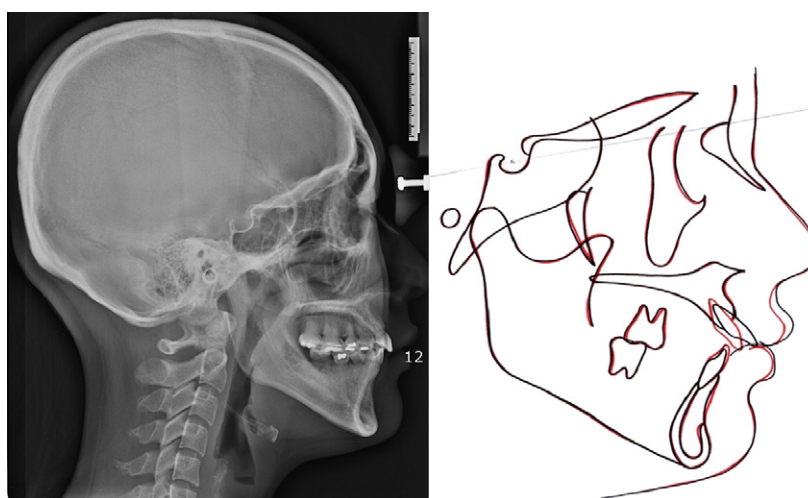


Fig 12. Posttreatment lateral cephalogram and its tracing. Retraction of the maxillary anterior teeth was accomplished, maintaining the sagittal position of the mandibular incisors.



Fig 13. Posttreatment panoramic radiograph.

REFERENCES

- Shields ED, Bixter D, El-Kafrawy AM. A proposed classification for hereditary human dentine defects with a description of a new entity. *Arch Oral Biol* 1973;18:543-53.
- Huth KC, Paschos E, Sagner T, Hickel R. Diagnostic features and pedodontic-orthodontic management in dentinogenesis imperfecta type II: a case report. *Int J Paediatr Dent* 2002;12:316-21.
- Kindelan J, Tobin M, Roberts-Harry D, Loukota RA. Orthodontic and orthognathic management of a patient with osteogenesis imperfecta and dentinogenesis imperfecta: a case report. *J Orthod* 2003;30:291-6.
- O'Connell AC, Marini JC. Evaluation of oral problems in an osteogenesis imperfecta population. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1999;87:189-96.
- Roberts E, Schour I. Hereditary opalescent dentine (dentinogenesis imperfecta). *Am J Orthod* 1939;25:267-76.
- Siar CH. Quantitative histological analysis of the human coronal dentine in dentinogenesis imperfecta types I and II. *Arch Oral Biol* 1986;31:387-90.
- Sunderland EP, Smith CJ. The teeth in osteogenesis and dentinogenesis imperfecta. *Br Dent J* 1980;149:287-9.
- Huber MA. Osteogenesis imperfecta. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2007;103:314-20.
- Battagel JM, Levenkind M. Dentinogenesis imperfecta: an interdisciplinary approach. *Br Dent J* 1988;165:329-31.
- Crowell MD. Dentinogenesis imperfecta: a case report. *Am J Orthod Dentofacial Orthop* 1998;113:367-71.
- Forbes GB, Taves DR, Smith FA, Kilpper RW. Bone mineral turnover in a patient with osteogenesis imperfecta estimated by fluoride excretion. *Calcif Tissue Res* 1978;25:283-7.
- Cole NL, Goldberg MH, Loftus M, Kwok V. Surgical management of patients with osteogenesis imperfecta. *J Oral Maxillofac Surg* 1982;40:578-84.
- Ormiston IW, Tideman H. Orthognathic surgery in osteogenesis imperfecta: a case report with management considerations. *J Craniomaxillofac Surg* 1995;23:261-5.
- Zola MB. Staged sinus augmentation and implant placement in a patient with osteogenesis imperfecta. *J Oral Maxillofac Surg* 2000;58:443-7.
- Lee YS, Ertel SK. Bone graft augmentation and dental implant treatment in a patient with osteogenesis imperfecta: review of the literature with a case report. *Implant Dent* 2003;12:291-3.
- Prabhu N, Duckmanton N, Stevenson AR, Cameron A. The placement of osseointegrated dental implants in a patient with type IV B osteogenesis imperfecta: a 9-year follow-up. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2007;103:349-54.

Table II. Cephalometric measurements before and after treatment

Measurement	Before treatment (°)	After treatment (°)	Change (°)
SNA	77	77	0
SNB	74	74	0
ANB	3	3	0
Gonial angle	129	129	0
FMA	33	33	0
FH to U1	130	113	-17
IMPA	85	86	1
Interincisal angle	109	127	16
NP to U1	12	8	4
NP to L1	3	3	0
Nasolabial angle	115	120	5