Dentinogenesis imperfecta: a challenge for root canal treatment — case report

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Dentinogenesis imperfecta (DI) is a hereditary disorder affecting both the deciduous and permanent dentition. Partially or totally obliterated root canals and the defective soft dentin structure present a challenge for root canal treatment. The following case report illustrates these difficulties and shares our experiences with root canal treatment of 2 teeth with apical pathosis in a 38-year-old female with type II DI. Tooth #34 exhibited an occasional sinus tract and partially obliterated root canal. After regular cleaning and shaping, the problem with the sinus tract was resolved, but thin dentinal walls resulted. Tooth #46 with a metal crown exhibited apical rarefactions of both the medial and distal roots. The pulp chamber and root canals were totally obliterated. Since the canal orifices could not be identified, mishaps such as perforation and canal deviation resulted. The present case report demonstrates that early recognition and diagnosis of DI are important. When a DI patient requires endodontic treatment, gentle instrumentation force should be applied to conventional root canal preparations. Treatment options like periapical surgery should be considered when the canal is completely obstructed. (J Dent Sci, 3(2):117-122, 2008)

Key words: dentinogenesis imperfecta, pulp obliteration, root canal treatment.

Dentinogenesis imperfecta (DI) is one of the most common genetic disorders affecting the structure of dentin. In DI, the teeth are discolored ranging from bluish-gray to yellowish-brown and translucent. Based on various clinical and radiographic features, Shields et al.1 subdivided DI into 3 groups. DI type I occurs in people with osteogenesis imperfecta (OI), a hereditary connective tissue disease characterized by bone fragility. Most types of OI are caused by mutations of genes encoding type I collagen. DI types II and III usually occur in people without other inherited disorders. DI type II, also called hereditary opalescent dentin, is the most common subgroup with an estimated incidence of 1 in 6000–8000 people in the US2. Both deciduous and permanent dentition is affected, appearing discolored with variable degrees of attrition. Radiographically, crowns are bulbous with cervical constrictions and the roots are short. The pulp chambers and root canals are usually obliterated due to dentin overproduction. There have been reports of a few families with type II DI having progressive hearing loss in addition to dental abnormalities. DI type III was first identified in a tri-racial isolate in Maryland, also known as the “Brandywine type”. Multiple pulp exposures and “shell teeth” (due to large pulp chambers and thin dentinal walls) are 2 characteristics used to distinguish DI type III from type II. Some researchers believe that DI types II and III, and dentin dysplasia type II (which is also a
hereditary dentin defect) are actually variant forms of the same disorder.\(^3\)

Formation of defective dentin in the structure and composition are the major anomalies of DI\(^4-6\). The enamel is usually reported to be normal, but is easy to chip or shear away from the dentin due to a lack of support from a normal scalloped dentin-enamel junction\(^6,7\). The early loss of enamel and poorly calcified dentin make these teeth prone to rapid wear. Teeth that undergo severe attrition may lead to pulp exposure and require endodontic treatment. However, narrowing of the root canal and the defective dentin structure present a challenge for root canal treatment. The purpose of this article is to report a clinical case of DI type II with more than 1 apical pathosis requiring endodontic treatment. Challenges and modifications of the treatment procedure for DI patients are also discussed.

**CASE PRESENTATION**

A 38-year-old female was referred to the Dental Department of Taipei Veterans General Hospital with a sporadic sinus tract on the lower left quadrant for more than 10 years. Her medical history was noncontributory, except for cryoglobulinemia. There were no skeletal or scleral abnormalities. Oral examinations revealed bluish-gray discoloration of the clinically visible teeth (Figure 1). The patient in recalling her family history remembered that 2 maternal cousins exhibited the similar dental problem. Short roots, narrowed pulp chambers, and totally obliterated root canals were striking radiographic findings of the posterior teeth (Figure 2). The clinical and radiographic findings supported a diagnosis of DI type II. Intraoral examinations and periapical radiographs also showed a 3-unit bridge on teeth #15–#17, single crowns on teeth #24, #26, #36, and #46, and deep fillings on teeth #25, #34, and #35. According to the patient, these full-coverage crown restorations were made because her teeth chipped easily. The root canals of the crowned teeth were totally obliterated, and no apical pathoses were noted. The exceptions were tooth #46 which had apical rarefaction and tooth #15 which exhibited periodontal ligament (PDL) widening. These teeth were asymptomatic, had probing depths of < 3 mm, and responded normally to percussion, palpation, and mobility tests. The findings of concern were apical radiolucencies on teeth #34 and #46.

A sinus tract on the buccal gingiva of tooth #34 was observed. Neither tooth #34 nor #46 exhibited tenderness to percussion or palpation. The periodontal probing depths were < 3 mm. The mobility was within normal limits. Tooth #34 had a buccal composite resin filling and showed no response to electric pulp testing. The periapical radiograph of tooth #34 showed a 6 × 6 mm radiolucency around the root apex and a faintly recognizable obliterated root canal. Tooth #46 had a metal crown with adequate margins, obliterated root canals, and 2 periapical radiolucencies on the mesial and distal roots sized 4 × 4 and 2 × 2 mm, respectively. The diagnosis of tooth #34 was necrotic pulp with supplicative apical periodontitis. Tooth #46 was diagnosed as necrotic pulp with chronic apical periodontitis. Nonsurgical endodontic treatment of teeth #34 and #46 was chosen after discussion with the patient.

In February 2007, the patient presented for root canal treatment on tooth #34. After rubber dam application and endodontic access opening, the root canal orifice was located after careful negotiation with an endodontic explorer (DG-16). The canal was cleaned and shaped to apical size 35 using hand instruments and copious irrigation with a 2.5% sodium hypochlorite solution. Calcium hydroxide/distilled water paste was placed as an intracanal medicament. Four days later, the patient called and reported pain and abscess formation over the buccal gingiva. Clindamycin (150 mg) every 6 hours for 5 days was prescribed. A week later, the patient presented without pain or abscess, but the sinus tract persisted. At this appointment, the root canal cleaning and shaping were regularly continued with Gates...
Glidden #2, #3, and #4 drills and hand instrumentation under rubber dam isolation. Calcium hydroxide/distilled water paste was placed in the root canal, and intermediate restorative material (IRM®, Dentsply) was placed to temporarily seal the access cavity. After 8 weeks, the tooth was asymptomatic, and the sinus tract had resolved. The root canal was obturated with gutta-percha and Roth's 801 sealer (Roth International, Chicago, IL, USA) using a lateral compaction technique. The master apical file was #50. However, the final radiograph showed thin dentinal walls due to over-enlargement of the root canal on the coronal 1/2 (Figure 3).

Eight weeks later, the patient presented for root canal treatment of tooth #46 and reported that her bridge on teeth #15–#17 had loosened. After drilling through the crown of tooth #46 under rubber dam isolation, severe calcification of the pulp chamber was detected. Attempts to negotiate under an operating microscope were made, but the floor map of the pulp chamber was difficult to recognize due to the absence of normal landmarks. Subsequently, a small supracrestal perforation of the distal cervical 1/3 was produced with a #15 K file. Calcium hydroxide
dressing, a cotton pellet, and a temporary restoration were put in place. The difficulties of nonsurgical endodontic treatment due to severe canal calcification and high risk of iatrogenic perforation due to the soft texture of tooth dentin were discussed with the patient. The bridge of the non-symptomatic teeth #15 and #17 was temporally recemented and placed under observation.

At the next appointment, tooth #46 remained asymptomatic. After finding a deviated pathway established for the MB canal, negotiation was stopped. The canal was obturated with gutta-percha and Roth's 801 sealer, followed by placement of an amalgam core (Figure 4).

The patient was asymptomatic and returned 6 months later. Teeth #34 and #46 responded with no tenderness to percussion or palpation tests. The periodontal probing depths were < 3 mm. Radiographic examinations showed significant improvement in the periapical lesion of tooth #34. No apparent radiographic changes in the other teeth were noted (Figure 5). Since the apical radiolucency of tooth #34 has not completely resolved and the lesion of tooth #46 persists, continued follow-up is imperative.

DISCUSSION

A striking radiographic feature of DI teeth is the partially or totally calcified pulp chamber and root canal. When the pulpal space is obstructed by overproduced dentin, it is difficult to recognize canal orifices and negotiate root canals. In addition to a thorough understanding of tooth anatomy, a dental operating microscope has proven to be a great asset in dealing with calcified canals. Some instruments (such as multiple-purpose probes and surgical-length contra-angle burs) and chelating agents are also suggested to help gain access and improve the possibility of negotiating calcified canals. However, the poorly mineralized dentin of DI teeth decreases its resistance to these instruments. Thus, sharp and efficient instruments might not be as helpful as they are on normal dentin. Procedural accidents, such as perforations and ledges, may be produced similar to the problems we encountered in treating tooth #46. Chelating agents are not advised in order to prevent further softening of the original defective dentin.

Obliterated root canals and soft dentin make orthograde conventional root canal treatment of DI-affected teeth an extremely arduous task, even for experienced and skilled endodontists. When proper instrumentation of a severely calcified canal is impossible, periapical surgery should always be considered for a tooth with persistent apical pathosis.

In DI-affected teeth, the microhardness of the dentin closely approximates that of cementum. Once the calcified root canal has been identified and negotiated, instrumentation should be performed very carefully. The goal of root canal treatment is to remove the infected dentin and preserve as much of the sound dentin as possible. However, the defective soft dentin is easily reduced, particularly when engine-driven rotary instruments are used. For this reason, we suggest rotary instruments be used with gentle force and as few times as required. Over-preparation of the dentinal wall will weaken the tooth structure and make teeth prone to root fracture.

Severe attrition can lead to pulp exposure as well as early loss of the vertical dimension. When the major part of the tooth is worn away, it is difficult to restore a short crown without a clearly identifiable root canal. Therefore, in a young patient with a highly progressive attrition rate, prophylactic crowns should be inserted on all molars and resin crowns on incisors to protect the teeth from further wear. With the exception of the early loss of tooth #16, this is the reason all first molars had been restored with metal crowns in the present case. Overlay dentures is another restorative treatment to establish the vertical dimension, restore function, and improve esthetics for patients suffering from extreme attrition.

Detachment of fixed prostheses seems to be a
RCT for dentinogenesis imperfecta teeth

concern with DI teeth. This situation also occurred on the bridge of teeth #15–#17 in this case. With superior bond strength, resin cement is suggested to overcome compromised retention problems such as in severely worn teeth. However, DI-affected dentin has irregular tubules, often with larger areas of uncalcified matrix, and contains up to 60% more water than do tubules in normal dentin. The wet environment and lack of a mineralized surface make bonding of resin theoretically more difficult. Still, clinical success in most patients is reported. Adhesive dentistry is not contraindicated in patients with DI. Regular follow-up of DI teeth after restoration is recommended to detect loose restorations as early as possible and prevent secondary caries.

Caries do not seem to be a more-frequent problem in DI-affected teeth. A proposed theory is that defective dentin exhibits some resistance due to the absence of dentinal tubules and to the inability of caries to develop on a surface where enamel is rapidly being lost to abrasion and fracture. However, teeth with rampant caries and a poor response to endodontic treatment have been reported. It is critical to provide proper oral hygiene instructions and regular follow-up in the maintenance phase.

Root canal treatment is difficult in DI teeth. The dentin is soft, and root canal instrumentation should be carefully performed so as not to remove too much of the tooth’s structure. Endodontic treatment is even more difficult when the root canal is totally obliterated by the continuous deposition of dentin. Surgical intervention is another treatment option when negotiation of the calcified canal is not possible. Early and correct diagnosis of DI is essential for optimal dental treatment.

REFERENCES


Figure 5. Six-month follow-up radiographs showing significant improvement in the periapical lesion of tooth #34. No obvious changes in the other teeth were noted.