Treatment of anterior open bite in a patient with dentinogenesis imperfecta

ABSTRACT

Dentinogenesis imperfecta (DGI) is a genetic disorder characterized by severe hypomineralization of dentin and an altered dentin structure. Teeth with this disease have an amber hue due to the brittle dentin, and exhibit frequent wear and chipping of the enamel and dentin due to insufficient adhesion to the superficial enamel. Herein, we report the successful orthodontic treatment of a patient with DGI. A 24-year-old woman had chief complaints of anterior open bite (AOB). She showed Class II and open bite skeletal discrepancy with excessive anterior facial height and lip incompetency due to mandibular backward rotation. Edgewise treatment with temporary anchorage devices were used to induce counterclockwise rotation of the mandible by intruding the molars and reducing the overjet by total arch distalization. Resultantly, the facial profile and occlusion showed substantial improvement without any noticeable side effect.

INTRODUCTION

Dentinogenesis Imperfecta (DGI) is an autosomal dominant disorder with an estimated prevalence ranging from 1/6000 to 1/8000 [1]. The disorder is classified into two types: Type I and Type II. Type I is associated with osteogenesis imperfecta (OI), which is caused by mutations in the collagen type 1 gene [2]. Type II is caused by a mutation in the dentin sialophosphoprotein (DSPP) gene [3,4]. DSPP governs the synthesis of non-collagenous proteins that play a pivotal role in the transformation of pre-dentin into mineralized dentin; therefore, its disruption can lead to brittle dentin [5].

In clinical examination, these teeth manifest an amber hue, exhibiting a spectrum that spans from grayish purple to yellowish-brown or a purplish-brown color change compared to that seen in non-affected individuals. Moreover, their dental morphology is characterized by bulbous contours and abbreviated roots. Radiographically, DGI is distinguished by pulp chambers that display a progressive obliteration because of the ongoing and irregular deposition of dentin by odontoblasts [6,7]. Insufficient adhesion of the enamel to the superficial dentinal layer results in its propensity to chip away from the dentin, thereby exposing the dentin and reducing the clinical crown height. Incomplete mineralization of the dentin also results in severe abrasion by occlusal forces.

The successful correction of an anterior open bite (AOB) is considered one of the most challenging treatments in the field of orthodontics [8]. Moreover, long term stability of the AOB cases relies on the selection of the proper therapeutic approach.

In recent years, multiple investigations have elucidated the efficacy of molar intrusion in managing individuals afflicted with AOB through the utilization of temporary skeletal anchorage devices (TSADs) [9-11]. This therapeutic approach has been demonstrated to result in consequential counterclockwise mandibular rotation, a decrease in anterior vertical facial height, anterior displacement of the mentum, and amelioration of the retrognathic facial profile [12].

It is also important to note that DGI poses challenges to orthodontic intervention because of its brittle dentin [13], and only a small number of orthodontic cases in individuals with DGI have been documented [14,15].

We report the successful orthodontic treatment of a woman with DGI who presented skeletal open bite and excessive anterior facial height, large overjet, and lip incompetency. The aim of this case report was to show the progress and outcome of an orthodontic treatment for a patient with DGI and discuss the specific attention that should be paid during such treatment.

DIAGNOSIS AND ETIOLOGY

A Japanese woman with DGI (aged 24 years and 3 months) first visited our hospital with a complaint of an AOB. Multiple restorations were done due to enamel chipping, dark blue-brown color of permanent teeth, and narrowed pulp cavity (Fig 1). The patient reported a history of early treatment with headgear at another hospital. However, this treatment was discontinued and, according to the patient, did not significantly improve the occlusion. At that time (age of 9 years and 10 months), intraoral photographs showed noticeable primary tooth abrasion.

An extraoral examination showed a symmetrical face, incompetent lip, deep labio-mental fold, and a proportionally long lower anterior facial height (Fig 1). An intraoral examination revealed AOB, large overjet, Angle Class II relationships, narrow maxillary arch, and mild crowding of the maxillary and mandibular incisors (Figs 1 and 2). The patient also had multiple restorations on the molars due to enamel chipping and dental caries. The maxillary dental midline almost coincided with the facial and mandibular dental midlines. Compared with Japanese norms, lateral cephalometric analysis of the patient showed a skeletal Class I relationship (ANB, 3.3°), a high mandibular plane angle (FMA, 40.0°) with increased gonial angle (138.8°), and increased anterior lower facial height (ANS-Me, 72.7 mm). The maxillary central incisor was labially inclined (U1-FH, 121.7°), while the mandibular central incisor was normally inclined (IMPA, 89.5°) (Table).

TREATMENT OBJECTIVES

Based on these findings, the patient was diagnosed with AOB and large overjet with high mandibular plane angle, proclined maxillary incisors, and incompetent lips. The treatment objectives were to correct the skeletal open bite and obtain ideal overbite, overjet, and facial profile. Special consideration was required for the condition of brittle dentin, such as selecting the treatment modality and materials for bonding.

TREATMENT ALTERNATIVES

The patient was presented with two treatment options.

1. Option 1 involved the extraction of four premolars and the use of a fixed appliance to create sufficient space to correct the incisor protrusion and crowding.

2. Option 2 was a non-extraction approach using a fixed appliance and TSADs in both arches to distalize the incisors.

The extraction of four premolars could result in more significant lip retraction compared to non-extraction treatments in nongrowing patients [16]. In this case, the facial profile did not require lip retraction and the need for incisor retraction was minimal. Conversely, the patient's retruded chin and AOB required attention. TSADs can intrude the buccal segment, which helps to bring the chin forward, thereby improving the anterior open bite and facial profile

[17]. For these reasons, we chose to use the second treatment plan in this case.

TREATMENT PROGRESS

First, the maxillary posterior segments were aligned with sectional arch wires in a 0.022 x 0.028-in preadjusted edgewise appliance. Simultaneously, TSADs (diameter, 1.6 mm; length, 6 mm; Dual-Top Anchors; Jeil Medical, Seoul, South Korea) were placed at the posterior mid-palatal suture and buccal alveolar bone between the roots of the maxillary second premolar and first molar. Four weeks after insertion, correction of the AOB was started. Intrusive force was applied to the maxillary molar segment from both the buccal and palatal sides. On the buccal side, the force was applied by connecting the TSADs to the buccal wire with an elastic chain. Similarly, on the palatal side, the force was applied using TSADs and a trans-palatal arch, also with an elastic chain (Figs 4 and 5). After intrusion of the maxillary molar segment, a 0.022 x 0.028-in preadjusted edgewise appliance was also placed into the maxillary anterior segment with a continuous 0.016-in heat-activated nickel-titanium wire in the maxillary arch (Fig 5). After leveling and alignment of the maxillary arch, a 0.019 x 0.025-in stainless steel wire was installed to distally move the maxillary arch using an elastic chain connected to the TSADs in buccal and long-hooks with the wire (Fig 5).

Similar to the maxillary treatment, the mandibular posterior segments were aligned and intruded with TSADs. These were implanted between the roots of the second premolar and first molar in the mandible and connected to the buccal wire with an elastic chain. A lingual arch was placed to prevent the buccal inclination of the molars. Detailing began with 0.019 x 0.025-in stainless steel wires. To prevent excessive labial tipping of the mandibular incisors, TSADs were connected to the edgewise appliance at the canines with a ligature wire (Fig 5). The total active treatment period lasted 33 months, during which the TSADs were used to intrude the molars for 12 months.

As a special consideration for the fragile dentin, we selected metal brackets that retained only via mechanical engagement, along with adhesives possessing plastic deformation capabilities due to the absence of inorganic fillers, to minimize the damage to the enamel surface when debonding (Super-Bond, SUN MEDICAL, Shiga, Japan). After removing the appliance, maxillary wrap-around retainers were placed.

TREATMENT RESULTS

Our treatment successfully intruded the maxillary molars allowing counterclockwise rotation of the mandible and therefore increasing the chin projection and reducing the lower facial height. Mutually protected occlusion was achieved with the improvement in AOB and excessive overjet by total maxillary arch distalization (Figs 6 and 9). Adequate overjet (2.5

mm) and overbite (2.5 mm) were obtained, and Class I canine and molar relationships were also achieved (Figs 6 and 7). Acceptable root paralleling without obvious root resorption was observed on a panoramic radiograph (Fig 8).

The cephalometric superimposition showed a decrease in mandibular plane angle by 1.4°. The maxillary first molars were intruded for approximately 3.0 mm toward the palatal plane and were distally moved by 1.5 mm. As a result, the maxillary incisors were lingually inclined by 3.3°. The mandibular central incisors were labially inclined by 2.7° to gain space to improve the crowding (Table).

The patient's occlusion and facial esthetics achieved with the orthodontic treatment were maintained over a 1-year retention period (Fig 10).

DISCUSSION

DGI was classified by Shield into three types: Shields type I DGI is intricately linked with osteogenesis imperfecta (OI), whereas Shields types II and III DGI are restricted to dentin involvement. Shields types II and III DGI have been suggested to be phenotypic variations of a single disease [18]. OI primarily manifests in the skeletal system, giving rise to skeletal fragility, deformities, and growth impairments [2,19]. In the craniofacial region, an underdeveloped nasomaxillary complex precipitates a counterclockwise rotation of the mandible, thereby engendering skeletal discrepancies between the jaws. These discrepancies manifest as dental malocclusions, including Class III malocclusion, anterior and posterior crossbite, and posterior openbite [20,21]. Our patient was diagnosed with Shields type II DGI as she exhibited no physical, facial, or occlusal manifestations associated with OI.

In patients with DGI, despite the appearance of a normal enamel, it is susceptible to detachment and fracturing under occlusal stress due to the fragile nature of the dentin [13,22]. Meticulous attention must be paid to the management of orthodontic devices as their removal can lead to enamel fractures. To reduce the risk of such fractures, the application of bands on all teeth and the utilization of glass ionomer cement for cement bonding were advocated [23]. However, recent reports have demonstrated successful bonding of orthodontic brackets [24]. In our case, favorable outcomes were achieved without enamel exfoliation by employing metal brackets that rely on mechanical engagement alone for bonding, along with adhesives possessing plastic deformation capabilities due to the absence of inorganic fillers.

Premolar extraction emerged as one of the optimal choices for addressing the AOB in our patient. Studies have shown successful orthodontic treatment with extractions in patients with DGI, without adverse root movement or resorption [24]. Successful orthognathic treatments have also been reported in DGI patients with skeletal discrepancies [25,26].

However, we deemed it unnecessary to create a space by extraction for minor crowding of both arches and for a harmonious profile. Consequently, a treatment plan was devised to rectify the AOB and crowding through molar intrusion and distal movement.

The use of the TSADs was feasible in the patient with type II DGI in this case. For the maxillary molar intrusion and distal movement, we used TSADs, which were stable enough to serve as a fixation source throughout the treatment duration. Successful cases have also been reported showing that TSADs in patients with type II DGI are sufficiently stable to be used throughout the treatment [24]. We thus consider that TSADs could be safely used in patients with type II DGI.

The orthodontic correction of AOB can be achieved through diverse mechanisms encompassing posterior teeth intrusion, anterior teeth extrusion, or a combination of them. When addressing the treatment of open bite, caution must be exercised when leveling the maxillary arch using a continuous archwire, as it may inadvertently lead to undesired incisor extrusion, particularly in adult patients. This can be attributed to the leveling of a maxillary arch featuring a biphasic occlusal plane, which is a prevalent characteristic of open bite [27]. Consequently, prior to the alignment of anterior teeth, segmental molar intrusion was implemented to counteract undesirable incisor extrusion. Additionally, TSADs were strategically positioned on both the buccal and palatal aspects, applying equal forces to minimize any buccopalatal side effects.

Conversely, it has been documented that even after achieving AOB improvement through a molar intrusion using TSADs, relapse may occur [11,28]. Our patient demonstrated satisfactory retention up to a 1-year post-retention period; however, long-term observation is imperative due to the heightened risk of relapse.

CONCLUSIONS

We described a successful orthodontic treatment with TSADs of a Japanese woman with DGI who showed a skeletal open bite, a skeletal Class I relationship, lip incompetence, long lower anterior facial height, and large overjet. As a result, her facial profile and occlusion improved without causing noticeable side effects such as enamel chipping, exfoliation, or tooth fracture. Our case report provides useful information for understanding the etiology, progress, and precautions that should be considered for the orthodontic treatment of patients with DGI.

ANB, the anteroposterior relation between the maxilla and mandible; SNA, the anteroposterior position of the maxilla relative to the anterior cranial base; SNB, the anteroposterior position of the mandible relative to the anterior cranial base; FMA, divergency of the mandibular plane relative to Frankfort horizontal plane; Gonial angle, the angle between the mandibular and ramus planes; U1-FH, maxillary central incisal to Frankfort horizontal plane; IMPA, mandibular central incisal to mandibular plane; IIA, the angle between maxillary and mandibular central incisal axes; N-Me, anterior facial height; N-ANS, upper anterior facial height; ANS-Me, lower anterior facial height.

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FIGURE LEGENDS

Figure 1. Pretreatment facial and intraoral photographs.

Figure 2. Pretreatment dental casts.

Figure 3. Pretreatment cephalograms and a panoramic radiograph.

Figure 4. A, Intraoral photographs taken during the intrusion of the maxillary molars. B, Schematic illustrations of the TSAD-induced intrusion of the maxillary molars.

Figure 5. Treatment progress during the correction of the anterior open bite: A, start of the sectional arch; B, start of intrusion; C, 16 months later; D, 26 months later.

Figure 6. Posttreatment facial and intraoral photographs.

Figure 7. Posttreatment dental casts.

Figure 8. Posttreatment cephalograms and a panoramic radiograph.

Figure 9. Superimposition of pretreatment (black) and posttreatment (red) tracings.

Figure 10. 1-year postretention facial and intraoral photographs.

Table I: Cephalometric measurements.