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#### CASE REPORT

# Orthodontic management of amelogenesis imperfecta: A case report

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#### **Key Clinical Message**

Amelogenesis imperfecta (AI) is a rare developmental anomaly characterized by poorly developed or absent tooth enamel, which complicates orthodontic treatment due to weak enamel-bracket bond strength. This case report presents a successful management of AI using fixed orthodontic appliances and prosthodontic rehabilitation.

#### Abstract

Amelogenesis imperfecta (AI) causes enamel defects, complicating oral hygiene, reducing masticatory function and lowering self-esteem. This case report details an 18-year-old female with AI who underwent fixed orthodontic treatment followed by prosthodontic rehabilitation. The multidisciplinary approach restored function and aesthetics, significantly improving her quality of life.

#### **KEYWORDS**

aesthetics, amelogenesis Imperfecta, case report, crowns, dentition, orthodontic appliance

#### 1 INTRODUCTION

Amelogenesis imperfecta (AI) is a rare developmental disorder characterized by defective or absent tooth enamel, affecting both primary and permanent dentition due to improper differentiation of ameloblasts.<sup>1,2</sup> While AI exclusively impacts enamel formation, it significantly reduces patients' quality of life by complicating oral hygiene maintenance, diminishing masticatory function, and lowering self-esteem.<sup>3</sup> Additional dental complications include rapid attrition, sensitivity, calculus deposition, and gingival recession.<sup>4</sup>

AI treatment strategies encompass restorative, prosthetic, periodontal, surgical, and orthodontic approaches. An interdisciplinary combination of these treatments has been shown to improve AI prognosis, particularly when initiated during the early permanent dentition stage. Orthodontists often encounter AI cases in the late mixed dentition stage and play a crucial role in aligning crowded teeth for subsequent prosthetic work. Management can involve fixed, removable, and clear aligner therapies, though fixed appliances pose challenges due to weak enamel. The conventional acid etch technique may lead to frequent bond failures and enamel chipping.<sup>5</sup> GIC-based resin cement is considered to enhance bracket retention on the enamel surface.<sup>6</sup> The debonding process should be modified to prevent enamel fracture. For aesthetic improvement, prosthetic rehabilitation is recommended.

This case report details the orthodontic treatment of an AI patient to align crowded dentition, followed by prosthetic rehabilitation using full or partial veneers made from various materials.

The rarity and novelty of this case lie in the fact that AI is an uncommon developmental anomaly, rarely encountered in routine practice. Orthodontic treatment,

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in this case, was particularly challenging and required a multidisciplinary approach, involving prosthodontists for crown and veneer placement, periodontists for crown lengthening, operative dentists for filling carious teeth, and orthodontists for tooth alignment to restore the patient's smile. Due to the defect in enamel formation, the bond strength between brackets and the tooth surface was weak, leading to multiple challenges throughout the treatment duration. Therefore, AI cases are rare and require special care during orthodontic treatment.

In this case, there was generalized malformed enamel and minimal clinical crown height in the lower molars. Restorations were performed to maintain the integrity of the teeth, making both banding and bonding particularly challenging.

# 2 | CASE HISTORY/ EXAMINATION

An 18-year-old female patient presented to the dental outpatient department at the College of Medical Sciences in Bharatpur, Nepal, with a chief complaint of yellowish tooth discoloration. She also reported decay in her lower teeth but had no history of sensitivity. The patient had experienced tooth discoloration since childhood, which was also present in her primary teeth. She expressed dissatisfaction with the unaesthetic appearance of her teeth. There was no relevant family history, and none of her family members exhibited similar dental anomalies.

Extraoral examination revealed no abnormalities. The patient presented with a consonant smile, though yellowish tooth discoloration was evident. Intraoral examination revealed a complete set of permanent dentitions with a bilateral Class I molar relationship and an end-on canine relationship on the right side. The patient exhibited a normal overbite with a reduced overjet. Crowding was observed in both the upper and lower anterior regions. Generalized discoloration was present, with severe attrition on the occlusal surfaces of the lower molars and slight chipping of the incisal edges of the lower incisors. Caries and chipped buccal enamel surfaces were noted on teeth 16, 26, 36, and 46. Oral hygiene was fair, with calculus and staining present in the lower anterior region.

# 3 | METHODS (DIFFERENTIAL DIAGNOSIS, INVESTIGATIONS, AND TREATMENT)

Diagnostic aids were utilized and analyzed. The orthopantomogram revealed thin enamel in the molars, with pulp chambers within normal limits. Based on the patient's history, radiographs, and clinical examination, a diagnosis of AI hypoplastic type with malocclusion was made.

A multidisciplinary treatment plan was devised, beginning with orthodontic alignment, followed by periodontal consultation for surgical crown lengthening, and concluding with prosthetic rehabilitation. Surgical crown lengthening was necessary due to the inadequacy of the clinical crowns for crown placement. The condition, treatment approach, and duration were thoroughly explained to the patient and her parents. Although the primary concerns were yellowish discoloration and decayed teeth, a detailed examination revealed malaligned teeth in both arches necessitating orthodontic treatment. The specifics of the orthodontic treatment were explained to the patient and her parents, who consented to the proposed plan. Informed and written consent was obtained, and the treatment commenced.

Caries and chipped buccal enamel surfaces on teeth 16, 26, 36, and 46 were restored using Glass Ionomer Cement (GIC). Orthodontic treatment was initiated for alignment and leveling. Oral prophylaxis was conducted to remove plaque and calculus, ensuring no interference with bonding. The enamel surface was etched with 37% phosphoric acid (Meta Etchant, Cheongju-si, Chungcheongbuk-do, Korea) for 30s, resulting in a white frosted appearance. The bonding agent was then applied and cured for 20s.

Orthodontic treatment commenced with the bonding of metal brackets (0.022 slots, Standard Edgewise 0.022×0.030 in.) (Morelli, Sorocaba, Brazil) using composite-based adhesives. Bands were placed on the upper molars, and bondable buccal tubes were applied to the lower molars. Initial 0.012" NiTi wires were ligated on both arches, progressing through 0.014", 0.016", and 0.016×0.022" NiTi wires, followed by 0.017×0.025" SS and 0.019×0.025" SS wires. Multiple bracket debondings occurred on the premolars, necessitating surface cleaning, re-etching, and re-bonding of the brackets. Leveling and alignment were completed within 4 months, with torquing and root paralleling requiring an additional 3 months. Upon achieving the desired objectives, debonding was performed. Debonding pliers were engaged occlusal and cervically on the bonding bases, applying peel force to break the bond between the bracket and resin (mechanical debonding process). Resin tags on the tooth surface were cleaned using a high-speed handpiece and tungsten carbide burs, ensuring a single-directional motion.

Figures 1A,B and 2A–J illustrate the pretreatment and post-orthodontic treatment appearances. Following orthodontic treatment, prosthetic rehabilitation was planned. All-ceramic laminates were fabricated for the upper anterior region and full-veneer all-ceramic crowns were placed on teeth 14 and 24. On a posed smile, both frontal and three-quarter views revealed visibility up to the upper first premolars. Therefore, considering aesthetics, function, and patient demand, the prosthesis was fabricated up to the upper first premolars.

In the mandibular arch, full-veneer crowns were fabricated for the anterior teeth, while all-metal crowns were placed on teeth 36 and 46. The upper teeth distal to the first premolars and the lower premolars and second molars remained intact, showing no signs of enamel loss or caries.

# 4 | CONCLUSION AND RESULTS (OUTCOME AND FOLLOW-UP)

The maloccluded teeth were successfully aligned, and prosthodontic rehabilitation was completed. The patient was scheduled for follow-up appointments and advised on preventive measures and home care to manage enamel loss and sensitivity. During these follow-ups, the condition of the retainer, the integrity of the prosthesis, potential relapse of the orthodontic treatment, and any chipping of the tooth structure were thoroughly evaluated.

The patient has been doing well during the two-year follow-up period, with no complications reported. The patient's post-prosthetic rehabilitation appearance at the two-year follow-up is shown in Figures 3 and 4A–E.

Malocclusion in AI hypoplastic type can be successfully treated using fixed orthodontic therapy. Upon completion of orthodontic treatment, further damage to the malformed tooth structure should be prevented by utilizing prosthetic crowns and veneers. Restoring decayed teeth in these cases as early as possible is crucial.

# 5 | DISCUSSION

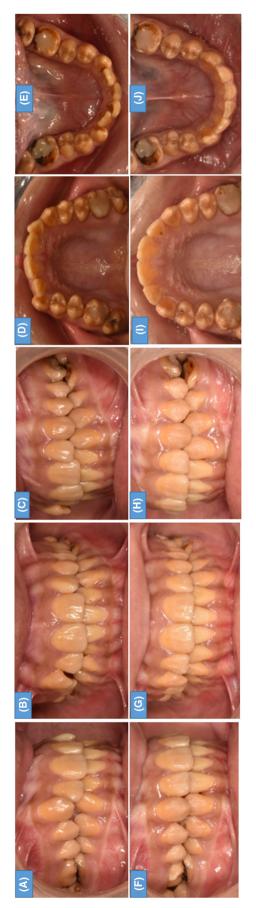
Amelogenesis imperfecta (AI) is a group of inherited disorders primarily affecting the structure of enamel. Witkop has classified AI into various types as shown in Table 1.<sup>1</sup> Patients with AI experience poor aesthetics, excessive tooth sensitivity, and compromised chewing function, often necessitating dental treatments from an early age. Due to malformed enamel, there is a high probability of enamel chipping, leading to exposure of the underlying dentin and pulp. The irreversible destruction of enamel often results in pain and tooth loss. Literature indicates that hydroxyapatite toothpaste effectively remineralizes enamel lesions and prevents further demineralization.<sup>7</sup> Atraumatic restorative technique (ART), Glass Ionomer Cement (GIC), and silver compounds are effective in such cases due to their minimally invasive technique, simplicity, and effectiveness in caries control.<sup>8</sup> In this case too before proceeding with fixed orthodontic treatment, all carious teeth were restored with GIC to prevent further enamel damage.

Achieving a successful outcome with fixed orthodontic therapy in cases of amelogenesis imperfecta (AI) is rare. Typically, affected individuals present with malformed crowns that are not conducive to the placement of brackets and bands, leading to frequent bracket debonding. Furthermore, inadequate clinical crown height in molars often complicates achieving adequate anchorage. These challenges were similarly encountered in our case. However, despite these obstacles, a favorable treatment outcome was achieved.

The general principles for managing AI are based on a comprehensive and timely approach, addressing the



FIGURE 1 (A)-Pretreatment extraoral smile photograph, (B) post orthodontic treatment extraoral smile photograph.





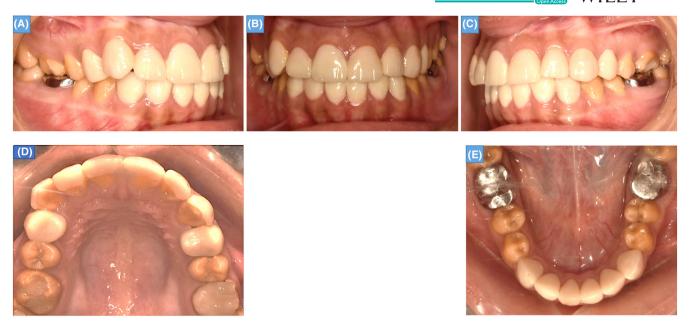


**FIGURE 3** Post-prosthetic rehabilitation extraoral smile photographs at 2-year follow-up.

clinical elements and the patient's demands and motivation.<sup>9</sup> Orthodontists must always start with the end in mind, aiming not just to achieve Andrews<sup>10</sup> six keys, a perfect occlusion, but to place teeth in positions that maximize the placement of restorations for function, aesthetics, and stability. A multidisciplinary team approach is required for proper treatment planning during initial and long-term phases. In this case, orthodontists, prosthodontists, periodontists and operative dentists were involved in the comprehensive treatment.

From an orthodontic standpoint, treatment options for AI include removable appliances, fixed appliances and clear aligner therapy. Removable appliances pose challenges in AI cases due to reduced crown height and lack of undercuts, complicating their placement and requiring greater patient cooperation.<sup>11</sup>

Fixed orthodontic treatment necessitates a wellprepared tooth surface and adequate bond strength of adhesive materials. The fragile enamel characteristic of AI patients poses difficulties during treatment and debonding processes.<sup>12</sup> Using a 37% etchant concentration with an etching duration of 30 s has demonstrated effective results. The literature suggests that glass ionomer cement-based adhesives should be utilized in AI



**FIGURE 4** (A, B, C, D, E) Prosthodontic rehabilitation intraoral photographs at 2-year follow-up, showing veneers in the upper anterior region, all-ceramic crowns on the upper first premolars and lower anteriors, and metal crowns on the lower first molars.

TABLE 1 Classification of amelogenesis imperfecta.

Type I: Hypoplastic IA: Hypoplastic: pitted autosomal dominant IB: Hypoplastic: local autosomal dominant IC: Hypoplastic: local autosomal Recessive ID: Hypoplastic: smooth autosomal dominant IE: Hypoplastic: smooth x-linked dominant IF: Hypoplastic: rough autosomal dominant IG: Enamel agenesis, autosomal recessive Type II: Hypomaturation IIA: Hypomaturation, pigmented autosomal recessive IIB: Hypomaturation, X-linked recessive IIC: Snow capped teeth, autosomal dominant Type III: Hypocalcified IIIA-autosomal dominant IIIB-autosomal recessive Type IV: Hypomaturation-hypoplastic with taurodontism IVA-Hypomaturation-hypoplastic with taurodontism, autosomal dominant IVB-Hypoplastic-hypomaturation with taurodontism,

autosomal dominant

cases to enhance appliance retention and mitigate further enamel demineralization, relying less on micro tag formation.<sup>13</sup> Gulnaz Marsan et al. treated a case of AI hypoplastic type in an 18-year-old female patient using fixed orthodontic therapy, achieving successful outcomes.<sup>14</sup> Similarly, Naomi Bechor et al. also treated a case of AI hypoplastic type in a 12-year-old female patient using fixed orthodontic therapy, with positive results.<sup>15</sup> This case was also treated with fixed orthodontic therapy using bondable composites. Clear aligners represent an alternative option for AI treatment, albeit with limited robust evidence supporting their efficacy. Existing literature primarily comprises case reports, case series, and expert opinions regarding their ability to predict orthodontic tooth movement.<sup>16</sup> Additionally, clear aligners are typically more expensive than fixed orthodontic therapy, limiting accessibility for some patients.

Existing literature suggests that all adhesive remnants should be removed using tungsten carbide burs in contraangled handpieces, employing light, single-directional strokes to avoid scratching or damaging the enamel surface.<sup>17</sup> In our case, we adhered to this protocol, consistent with the recommendations in the literature.

All ceramic veneers were fabricated for the upper anterior teeth due to sufficient tooth structure and the advantage of requiring less tooth reduction while providing optimal aesthetics. Full-ceramic crowns were fabricated for the first premolars (teeth 14 and 24) because of their critical role in mastication and the forces they endure. Full veneer crowns were also made for the lower anterior teeth, as existing literature suggests that partial veneers are less retentive due to their smaller dimensions.<sup>18</sup> Allmetal crowns were made for teeth 36 and 46 due to the lower clinical crown height, in line with literature suggesting a high risk of pulp exposure in young permanent teeth with porcelain-fused-to-metal (PFM) crowns.<sup>19</sup>

Existing literature suggests that to prevent further damage and sensitivity due to enamel loss in non-restored teeth, patient counseling on hygiene practices (e.g., type and hardness of toothbrush, brushing before or after meals), diet (e.g., frequency of food and acidic beverage intake), and other harmful habits is crucial.<sup>20</sup> Additionally, the home use of fluoridated products, as well as potassium nitrate and strontium acetate with fluoride in dentifrices and mouthwashes, can benefit patients by reducing sensitivity and preventing caries.<sup>21</sup> In the present case, similar precautions were advised to the patient.

Gulnaz Marsan et al. treated a case of AI hypoplastic type in an 18-year-old female patient, achieving bilaterally Class I molar and canine relationships, normal overjet, overbite, and functional occlusion.<sup>14</sup> Naomi Bechor et al. also treated a case of AI hypoplastic type in a 12-year-old female patient, achieving similar outcomes.<sup>15</sup> This case also concluded with the same successful results consistent with the literature.

Ronald D. Venezie et al. suggest that sodium hypochlorite (NaOCl) is an effective protein denaturant capable of removing excess enamel protein. Pretreating AI enamel with sodium hypochlorite enhances the accessibility of enamel crystals to the etching solution, yielding a more clinically favorable etched surface. They pretreated teeth with 5% NaOCl for 1 min in cases of severely affected hypocalcified type AI and achieved excellent results.<sup>22</sup> In this case, we did not pretreat the enamel with NaOCl; instead, we directly applied the etchant and bonding agent and achieved the expected outcome.

# **Patient's Perspective**

The patient expressed high satisfaction as a result of the successful treatment, which was accomplished without any complications.

#### AUTHOR CONTRIBUTIONS

Kanistika Jha: Conceptualization; data curation; formal analysis; funding acquisition; investigation; methodology; project administration; resources; software; supervision; validation; visualization; writing – original draft; writing – review and editing. Manoj Adhikari: Conceptualization; data curation; formal analysis; funding acquisition; investigation; methodology; project administration; resources; software; supervision; validation; visualization; writing – original draft; writing – review and editing. Suraksha Shrestha: Conceptualization; data curation; formal analysis; resources; supervision; validation; writing – original draft; writing – review and editing. Anisha Pandey: Conceptualization; investigation; writing – original draft; writing – review and editing.

### FUNDING INFORMATION

The present case study has received no financial aid.

## CONFLICT OF INTEREST STATEMENT

Authors have stated explicitly that no potential conflict of interest has been reported in connection with this article.

#### DATA AVAILABILITY STATEMENT

No data was generated or analyzed in the present research.

#### ETHICAL APPROVAL

It is our routine standard surgical procedure so ethical clearance was not required from the institution's ethics committee, College of Medical Sciences, Bharatpur, Chitwan, Nepal.

#### CONSENT

Written informed consent was obtained from the patient to publish this case report in accordance with the journal's patient consent policy.

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# REFERENCES

- Witkop C J Jr. Amelogenesis imperfecta, dentinogenesis imperfecta, and dentin dysplasia revisited: problems in classification. *J Oral Pathol* 1988; 17: 547–553.
- Seymen F, Kiziltan B. Amelogenesis imperfecta: a scanning electron microscopic and histopathologic study. J Clin Pediatr Dent. 2002;26:327-335.
- Coffield KD, Phillips C, Brady M, Roberts MW, Strauss RP, Wright JT. The psychosocial impact of developmental dental defects in people with hereditary Amelogenesis imperfecta. J Am Dent Assoc. 2005;136(5):620-630.
- 4. Poulsen S, Gjorup H, Haubek D, et al. Amelogenesis imperfecta systemic literature review of associated dental and orofacial abnormalities and their impact on patients. *Acta Odontol Scand*. 2008;66(4):193-199.
- 5. Seow W, Amaratunge A. The effects of acid-etching on the enamel from different clinical variants of amelogenesis imperfecta: an SEM study. *Pediatr Dent*. 1998;20:37-42.
- Saroğlu I, Aras S, Oztaş D. Effect of deproteinization on composite bond strength in hypocalcified amelogenesis imperfecta. *Oral Dis.* 2006;12:305-308.
- O'Hagan-Wong K, Enax J, Meyer F, Ganss B. The use of hydroxyapatite toothpaste to prevent dental caries. *Odontology*. 2022;110(2):223-230.
- Torres PJ, Phan HT, Bojorquez AK, Garcia-Godoy F, Pinzon LM. Minimally invasive techniques used for caries management in dentistry. *A Review J Clin Pediatr Dent*. 2021;45(4):224-232.
- Seow WK. Clinical diagnosis and management strategies of Amelogenesis imperfecta variants. *Pediatr Dent*. 1993;15(6):384-393.
- Andrews L. The six keys to normal occlusion. Am J Orthod. 1972;62:296-309.

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- 11. Meeran NA. Iatrogenic possibilities of orthodontic treatment and modalities of prevention. *J Orthod Sci.* 2013;2(3):73-86.
- Arkutu N, Gadhia K, McDonald S, Malik K, Currie L. Amelogenesis imperfecta: the orthodontic perspective. *Br Dent J*. 2012;212(10):485-489.
- Hiraishi N, Yiu CK, King NM. Effect of acid etching time on bond strength of an etch-and-rinse adhesive to primary tooth dentine affected by amelogenesis imperfecta. *Int J Paediatr Dent.* 2008;18:224-230.
- Marşan G, Aksu IS, Kurt H, Kuvat SV, Cura N. Interdisciplinary treatment of a patient with amelogenesis imperfecta, a skeletal class III relationship, and an anterior open bite. *World J Orthod*. 2010;11(3):284-290.
- 15. Bechor N, Finkelstein T, Shapira Y, Shpack N. Conservative orthodontic treatment for skeletal open bite associated with amelogenesis imperfecta. *J Dent Child*. 2014;81(2):96-102.
- 16. Sawan NM. Clear aligners in patients with amelogenesis and dentinogenesis imperfecta. *Int J Dent.* 2021;2021(1):7343094.
- 17. Oliver RG. The effect of different methods of bracket removal on the amount of residual adhesive. *Am J Orthod Dentofacial Orthop*. 1988;93(3):196-200.
- Akin H, Tasveren S, Yeler DY. Interdisciplinary approach to treating a patient with Amelogenesis imperfecta: a clinical report. *J Esthet Restor Dent*. 2007;19(3):131-136.

- 19. Chen CF, Hu JC, Bresciani E, Peters MC, Estrella MR. Treatment considerations for patient with Amelogenesis Imperfecta: a review. *Braz Dent Sci.* 2013;16(4):7-18.
- Gillam DG, Orchardson R. Advances in the treatment of root dentine sensitivity: mechanisms and treatment principles. *Endod Top.* 2006;13(1):13-33.
- 21. Pashley DH. Dentin permeability, dentin sensitivity, and treatment through tubule occlusion. *J Endod*. 1986;12(10):465-474.
- 22. Venezie RD, Vadiakas G, Christensen JR, Wright JT. Enamel pretreatment with sodium hypochlorite to enhance bonding in hypocalcified amelogenesis imperfecta: case report and SEM analysis. *Pediatr Dent.* 1994;16:433-436.

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