

Fixed Rehabilitation of an ACP PDI Class III Patient with Amelogenesis Imperfecta

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Abstract

This clinical report describes the oral rehabilitation of a 19-year-old woman diagnosed with hypoplastic-type amelogenesis imperfecta (AI). She presented with discolored and mutilated teeth, missing teeth, anterior open bite, and posterior short crowns. She was classified as an American College of Prosthodontists Prosthodontic Diagnostic Index (ACP PDI) class III patient. The aim of treatment was to restore esthetics and improve masticatory function. The patient's esthetic and functional expectations were successfully attained with porcelain laminate veneers and all-ceramic and metal-ceramic restorations after extensive crown lengthening. The patient was regularly recalled during the postoperative period. Loss of retention of one all-ceramic crown and minimal recession of the gingiva of one laminate veneer were noted at 4-year recall.

Amelogenesis imperfecta (AI) is a genetically and clinically diverse group of hereditary disorders that primarily affect the quantity, structure, and composition of enamel.¹ The prevalence of AI varies among populations, ranging from 1:14,000 in the United States¹ to 1:4000 in Sweden.² The inheritance pattern may be autosomal dominant, autosomal recessive, or X-linked.

Of the various classification systems proposed for AI types,³ Witkop's classification system based on analysis of pedigree and phenotype is the most frequently cited. According to the Witkop classification system, there are four main forms of AI: hypoplastic, hypocalcified, hypomatured, and AI with tarudontism.¹ Hypoplastic AI is characterized by defects in the secretary process of ameloblasts that result in thin or pitted enamel. which may be normal or altered in structure or composition. Hypocalcified AI results from an inability of crystallites to properly nucleate, which causes abnormal crystallite growth and decreased mineral content in enamel. Hypomature AI is caused by abnormal processing of the matrix proteins during maturation, resulting from either abnormal cleavage of enamel matrix proteins or abnormal proteinase activity.4 Clinical presentation of AI varies considerably among the different AI types. While enamel in hypoplastic AI cases generally appears pitted, thin, and tinged with a vellow-brown color, it has a hard texture. The enamel is soft, opaque, and mottled white, yellow, or brown in hypomature AI. In hypocalcified AI, enamel is generally abraded and easily detachable from the underlying dentin.^{5,6} The teeth in AI patients can be severely worn and may lack mesial or distal contact. Despite the presence of defective enamel, affected teeth often remain relatively caries-free.⁷⁻⁹

Although AI primarily affects enamel formation, a variety of nonenamel manifestations may also be present, such as delayed eruption, congenitally missing teeth, crown resorption, root resorption, pulp calcification, and gingival hyperplasia.⁹⁻¹⁵ It is not known whether these abnormalities are also the product of a genetic amelogenesis abnormality or are only associated effects of a distinct etiology.⁴ AI has been shown to be associated with other craniofacial features. For example, AI has been associated with other craniofacial features. For example, AI has been associated with a more constricted maxillary arch (omega-shaped arch), a reversed curve of Spee, a vertical growth pattern, and skeletal open bite with or without the clinical presence of anterior space resulting in occlusal instability.¹⁶⁻¹⁹ Though a skeletal open bite is more prevalent in people with AI than in the general population,¹⁹ the mechanism underlying this link has not been resolved.

The primary clinical problems present in AI patients, regardless of subtype, are unsatisfactory esthetics, dental sensitivity, and loss of occlusal vertical dimension (OVD) due to the rapid wearing of dentition.^{5,20} There is no standard formula of care for successful treatment. While some patients may be treated with more conservative methods,^{12,21-23} it has been a challenge to achieve an esthetic restoration together with a functional and stable occlusion in the most severe cases.^{8,9,11,24-26} This clinical report describes the fixed rehabilitation of an American College of Prosthodontists Prosthodontic Diagnostic Index (ACP PDI) class III young adult hypoplastic AI patient.²⁷

Chief complaint

A 19-year-old white woman was referred to the Prosthodontics Department at Tehran University of Medical Science by her general dentist. She sought to improve the appearance of her anterior teeth and her chewing ability (Fig 1). She did not complain about tooth sensitivity, but diet analysis revealed that she avoided eating some foods such as citrus fruits.

Past medical and dental history

The patient had a history of thyroid hyperfunction, which was controlled by medication. No other remarkable findings were identified in her medical record. According to the patient, none of her first-degree family members (mother, father, a sister, and a brother) exhibited the same dental problems. She had not received any prior dental treatment despite having seen a dentist. She adhered to a regular daily brushing routine.

Extraoral examination

The patient showed no facial asymmetry and had competent lips. No muscle tenderness or palpable lymph nodes were detected. No signs or symptoms of joint disorder, including pain, limited range in jaw opening, or clicking were observed. No problematic oral habits, such as lip sucking or misuse of foreign objects, were revealed by the interview and physical examination.

Intraoral examination

The patient's enamel was yellow-brown, but was hard and exhibited no signs of detachment by an explorer. Tooth surfaces were found to be dull and rough; however, the cervical region of the teeth had a normal clinical appearance (Fig 2). The incisal edges were thin, and the cuspal structures were aberrant. There were short clinical crowns, especially in the posterior regions. Other findings included anterior tongue thrust, caries on tooth #27, and simultaneous pain on teeth #14 and 15. Teeth #2 and 3 were completely covered by gingival overgrowth, and cusp imprints from opposing teeth were observed on the gingival tissues. The patient's oral hygiene was generally poor, as evidenced by 21% bleeding on probing and 32% plaque index findings. No pocket deeper than 4 mm was recorded. Esthetic evaluation revealed a "gummy smile" and asymmetry of gingival contours in the maxillary anterior sextant. A phonetic evaluation yielded normal findings. All other tissues appeared to be normal and healthy.

Occlusal findings

In the maximum intercuspal position, there was an approximately 3-mm space between the anterior incisors. There was no occlusal contact mesial to the second premolars. Teeth #31 and 32 were in contact with the gingival pad on the upper arch. Teeth #17 and 18 had unstable occlusal contacts. Anterior disclusion occurred upon excursion movements by posterior segments. There was no interference on balancing sides; however, the most posterior parts of the maxillary and mandibular arches made contact during protrusive jaw movement. The interocclusal distance measured at the premolar region during physiological rest was 2 mm. It was not possible to determine the molar relationship, and a class III canine relationship was observed.

Radiographic findings

A panoramic radiograph revealed a thin layer of enamel along the top of most erupted teeth (Fig 3). The crowns of teeth #2 and 3 were resorbed.^{10,28} The roots of all teeth appeared to be normal in size and shape; however, the pulp chambers and root canals were abnormally large. Several closely approximated roots were also evident. Pulpal involvement was found on teeth #2, 3, 14, and 15. Cephalometric analysis revealed a class I skeletal relationship, a vertical growth pattern, and skeletal open bite (Sella-Nasion-A point [SNA] = 81°, Sella-Nasion-B point [SNB] = 78°, A point-Nasion-B point [ANB] = 3°, maxillarymandibular plane angle [MMP] = 36°, Gonial angle = 137°, and facial index = 60%).¹⁶

Diagnosis

The patient was diagnosed with hypoplastic-type AI the and designated as ACP PDI class III for partial edentulism.²⁷ Her problems were listed as follows: generalized mild gingivitis; unerupted teeth #2 and 3; pulpal involvement of teeth #14, 15, and 27; short clinical crowns in posterior segments; insufficient interocclusal space in molar regions; large pulp chambers; anterior open bite; occlusion instability; and esthetic problems due to discolored teeth, gummy smile, and uneven gingival margins.

Treatment plan

A treatment plan was developed with the aims of pain control for teeth #14 and 15; general preventive care and improvement in oral hygiene, caries removal, and root canal therapy for teeth #15 and 27; orthodontic consultation for improving the occlusal plane and managing the anterior open bite; periodontal correction of the gingival contour in the anterior sextant; and crown lengthening in the posterior regions. Esthetic and occlusal evaluations, including diagnostic wax-up on mounted casts, were included in the treatment plan.

The prosthodontic treatment planning was as follows: preparing the anterior teeth for porcelain veneers and establishment of anterior guidance, fabricating cast dowels, cores, and crowns for teeth #2, 3, and 15. A metal-ceramic fixed partial denture (FPD) was included for replacing tooth #14 and all-ceramic crowns on teeth #4, 5, 11, 20, 21, 28, and 29.

Treatment

The proposed treatment plan was discussed with the patient and her family. The patient emphasized the importance of improving her appearance. It was explained to her and her parents that the functional reconstruction was at least as important as esthetic improvement to protect the dentition from being further worn or damaged. The patient consented to the recommended mouth reconstruction, but refused any orthodontic treatments due to the financial burden.

Maxillary and mandibular complete arch primary impressions were made using a heavy and light body vinylpolysiloxane impression material (Imprint, 3M ESPE, Seefeld, Germany). Two sets of diagnostic casts were made by pouring the impressions twice with a type III dental stone (Fuji Rock, GC Dental-Corp., Tokyo, Japan). One cast set was used for the diagnostic wax-up, and the other was saved for other consultations and patient records. The casts were mounted on a semi-adjustable articulator (Denar Mark II, Teledyne Water Pic, Fort Collins, CO) using a facebow transfer (Denar Slidematic, Teledyne Water Pic) and the centric relation record.

A flat-end plane anterior deprogrammer was fabricated in the mouth with autopolymerizing resin (GC Pattern Resin, GC Dental Products Corp.) and was left in place for 30 minutes to erase muscle engrams. A centric relation record was taken according to Dawson's bimanual technique.²⁹ There were no differences between centric relation and maximum intercuspation. The articulator was programmed based on excursive records. The diagnostic wax-up served as a model to guide esthetic interventions, including closure of the space between the anterior teeth and development of a harmonic and functional anatomy (Fig 4). A Broadrick Occlusal Plane Analyzer was used to determine the occlusal plane and curve of Spee.³⁰ The occlusal plane analysis indicated that no correction of mandibular molars was required; thus, as they were not esthetically involved or decayed, it was decided not to restore them.

The patient's oral hygiene program started with scaling and prophylaxis, a 0.12% chlorhexedine gluconate (Darupakhsh, Karaj, Iran) oral rinse twice a day, and oral hygiene instructions. After the patient demonstrated adherence to the recommended oral health care program for 1 week, periodontal surgical procedures were planned to improve the gingival contour in the esthetic zone, reduce the amount of gingival display during smiling, improve the height of the posterior clinical crowns, and uncover teeth #2 and 3 (Fig 5). Crown lengthening was conducted over three sessions. Bone reduction in the upper quadrants was required to establish the needed biologic width. Tooth #14 was extracted during the bone reduction session. To gain sufficient height, it was also necessary to remove the mesiobuccal root of the maxillary second molar. The tooth proximity of the first and second molars was concomitantly corrected by this surgery. Extraction of tooth #14 and uncovering of teeth #2 and 3 resulted in reduction of the vertical dimension and the anterior open space by 1 mm, which enhanced the esthetic outcome, but had a limited impact on the prosthodontic prognosis of the maxillary molars due to insufficient vertical space.

After 4 weeks, endodontic therapy was completed on teeth #2, 3, 15, and 27. Two weeks later, the anterior teeth were prepared for porcelain laminate veneers. The amount of re-

duction was guided by the silicone index of the diagnostic wax-up, which was also used for fabricating provisional composite veneers (Protemp II, 3M ESPE). A maximum reduction of 0.5 mm on the facial surfaces of the anterior teeth using a self-limiting depth-cutting bur (Model 834-016, Brasseler USA, Savannah, GA) followed by a round-end tapered diamond (Model 856-016, Brasseler USA) was needed to reduce the facial surfaces and hence create a chamfer cervical finish line. The teeth showed shallow enamel defects limited to twothirds of the tooth surface area. It was confirmed that the enamel defects were sufficiently shallow to proceed with the plan for placement of porcelain laminate veneers, thereby avoiding extensive removal of sound tooth structure. The amount of incisal reduction was determined by the amount of interocclusal space. The incisal edges of teeth #7 and 10 were chipped off during preparation, which was attributed to their insufficient thickness. After completing the preparation, the composite interim restorations were bonded with a drop of unfilled resin (Heliobond, Vivadent, Schaan, Leichtenstein) and applied to the prepared teeth, which had been etched with 37% phosphoric acid (Total Etch, Vivadent) on a small area in the middle of the labial surface.

Before bonding the interim restorations, definitive maxillary and mandibular impressions were obtained in vinylpolysiloxane impression material (Imprint), and definitive casts were formed for the fabrication of porcelain laminate veneers (Colorologic, Ceramco, Burlington, NJ). The fit of each veneer was evaluated intraorally and then the veneers were silanized (Monobond-S, Vivadent). Enamel surfaces were etched for 40 seconds with 37% phosphoric acid (Total Etch). The restorations were then bonded with a dual polymerization resin cement (Variolink II, Vivadent), a dental adhesive system (Syntac Primer and Syntac Adhesive, Vivadent), and a bonding material (Heliobond) with minimum occlusal correction.

Cast posts and cores were fabricated from silver palladium alloys (Degussa-Huls, Hanau, Germany) using an acrylic resin pattern (GC Pattern Resin) formed in the mouth on teeth #2, 3, and 15. The cast posts and cores were adjusted as needed and cemented with zinc phosphate cement (Harvard Cement, Richter & Hoffmann, Berlin, Germany). An anterior jig was formed using an acrylic resin pattern (GC Pattern Resin) to maintain OVD during preparation of the posterior teeth. Teeth #2 and 3 were prepared for a full metal restoration, while teeth #13 and 15 were prepared for metal-ceramic crowns and an FPD with metal occlusal surfaces. Gold occlusal surfaces were selected due to inadequate occlusal vertical space, insufficient crown heights, and their property of inducing relatively low wear on opposing dentition.^{31,32} Margins were prepared with chamfer and an intrasulcular deep chamfer. Teeth #4, 5, 12, 21, 20, 28, and 29 were prepared for all-ceramic crowns with a round shoulder finish line placed at the gingival level. Complete arch definitive impressions were made with vinylpolysiloxane (Imprint). All prepared teeth were temporized with laboratory-processed provisional crowns, which were fabricated with the aid of the diagnostic wax-up. The provisional crowns were relined (Tempron, GC Dental Products Corp.) and luted with a noneugenol cement (RelyX Arc, 3M ESPE). The occlusion was adjusted on provisional crowns to establish a mutually protected scheme in the mouth. An irreversible hydrocolloid impression (CA 37 Cavex,



Figure 1 Pretreatment appearance of patient.



Figure 2 Pretreatment frontal view.



Figure 3 Pretreatment panoramic radiograph showing a thin layer of enamel on the most erupted teeth and unerupted maxillary molars.



Figure 4 Diagnostic wax-up.

Haarlem, The Netherlands) was obtained to make casts to serve as a guide for the fabrication of definitive crowns. The metal substructures of the metal-ceramic restorations [Degubond G (Au 86%, Pt 10.4%, Rh 1.6%), Degussa-Huls] and all-ceramic copings (IPS Empress II, Ivoclar, Schaan, Leichtenstein) were preliminarily evaluated in place to examine the marginal fit and to finalize the shade selection for all-ceramic restorations. Trial insertion of definitive restorations before glazing was also performed to evaluate occlusion.







Figure 5 (A, B) The amount of crown lengthening and gingiva recontouring were marked on the diagnostic casts.



Figure 6 A 3-year post-treatment review of the restorations: (A) frontal view; (B) maxillary occlusal view; (C) mandibular occlusal view; (D) right working side; (E) left working side.

The completed metal-ceramic restorations were cemented with glass ionomer cement (Ketac Cem, 3M ESPE). The interior surfaces of the all-ceramic restorations were etched with 4.9% hydrofluoric acid (IPS Ceramic gel, Vivadent) for 20 seconds and silanized (Monobond-S). The restorations were bonded with dual polymerization resin cement (Variolink II). The mutually protected occlusal scheme was preserved for this patient to allow for relatively even distribution and less stress of forces during excursive movements (Fig 6). There was no interference

observed in the anterior guidance of posterior teeth during lateral and protrusive movements. The patient reported that she was satisfied and comfortable with the treatment result (Fig 7). The restorations have since been in use for 4 years with no serious complications, but loss of retention of the all-ceramic crown on tooth # 5, which was recemented following the same bonding protocol, and a minimal recession on the gingiva of tooth #8. The patient showed no intention of replacing the laminate veneer of tooth #8.



Figure 7 Facial appearance of the patient (3 years post-treatment).

Discussion

There have been several clinical reports describing restoration treatments in AI patients, mostly in children and adolescents. These patients are sometimes psychologically affected by poor esthetics and may require extensive restorative treatment.^{15,20,22} Treatment may decrease social interaction-related anxiety and can substantially improve patients' quality of life.^{20,33} AI patients have previously been treated with multiple extractions followed by the construction of an overdenture or complete dentures.²¹ Such aggressive treatment courses have become unacceptable by modern dental professionals in light of advances made in the field of esthetic dentistry, especially with respect to tooth bonding systems and adhesive techniques. Several factors may influence the outcome of restorative treatments involving acid-etching and bonding of teeth affected by AI. For example, etch patterns of enamel may be altered by clinical variants of AI and thus not produce a good match to normal enamel.³⁴ Additionally, the morphological pattern of dentin in hypocalcified AI is relatively similar to sclerotic dentin, which responds to acid conditioning differently than normal dentin.35,36

Newer dentin-bonding systems provide more reliable bonding to dentin than did earlier systems. Although the newer systems also infiltrate the enamel prism more effectively, they may provide more durable dentin bonding than traditional methods of bonding to abnormal enamel.¹⁵ Despite severe enamel abnormalities, successful bonding of porcelain restorations could be achieved, and there were few adhesion complications in this case and several previously reported cases.^{12,22-26,30}

The decision as to whether to preserve an enamel layer and use adhesive restorations or to completely remove the enamel and use complete coverage crowns depends on the extension and depth of the patient's enamel lesions. 23,26 In the present case it was determined that the enamel should be preserved based upon observation of clinically normal enamel during preparation of the anterior teeth. Clearly, the precise composition of materials and methods selected depend upon the individual patient's situation. In the present case, porcelain laminate veneers offered a conservative option for anterior teeth to concomitantly restore anterior space and produce a marked esthetic improvement. Meanwhile use of all-ceramic crowns in the premolar region provided esthetic improvement, functional strength, and biocompatibility. Metal-ceramic restorations with a metal occlusal surface were selected for the posterior maxillary teeth, because they required minimum reduction. Furthermore, precious metal surfaces are relatively compatible with opposing mandibular molars, which were left intact in this case. 31,32

An interesting observation in the present case was the crown resorption of unerupted teeth. Although this phenomenon has been described in isolated cases, there has been little research addressing the cause. Crown resorption has been described as an irregularly distributed moth-eaten coronal area visible on unerupted teeth.¹⁰ Pre-eruptive crown resorption in autosomal recessive AI was investigated recently.²⁸ The authors described it as a rare, asymptomatic, clinical entity discovered only as an incidental radiograph finding with unclear etiology. The resorption was progressive due to asymptomatic true external recompliance. Early intervention to expose the involved teeth is recommended, as delayed treatment by general dentists in the present case resulted in further complications in the treatment procedures.

Conclusions

Designing an optimum treatment plan to achieve esthetic improvement and functional rehabilitation in AI patients can be challenging. Poor esthetics, worn dentition, and interocclusal space problems (both insufficient space and open bite) complicate the treatment course of AI patients. This clinical report describes esthetic and functional rehabilitation of a case of hypoplastic AI. The present treatment plan was developed specifically for the circumstances of this particular patient. Allceramic materials and adhesive techniques were used with no serious complications for a 3-year follow-up period. Early treatment of AI patients can prevent progressive damage of dentition and the psychological impact of the condition. Controlled retrospective studies of the prosthodontic management of AI patients and more clinical cases with an ample follow-up period are needed to increase our knowledge of treatment outcomes in patients with AI.

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