

Case Report

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Prosthetic Reconstruction of a Patient with Amelogenesis Imperfecta: A Two-Year Follow-up Evaluation

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Abstract

Background: Amelogenesis imperfecta (AI) has been described as a heredity disorder affecting dental enamel formation. **Case Report:** This report represents a step-by-step treatment plan for the dental reconstruction of a patient suffering from AI complaining about esthetics and function. The treatment sequences in the rehabilitation in the present case included performing orthognathic and periodontal surgery, using CAD-CAM provisional restorations during the healing time, and preparing full-ceramic restorations (zirconia restorations). CAD-CAM provisional restorations reduce the risk of pulp sensitivity and ensure adequate strength during the healing process.

Discussion: Oral manifestations of patients with AI include gingivitis, tooth sensitivity, deposition of calculus, anterior open bite, excessive wear and decrease of occlusal vertical dimensions, poor esthetics, loss of proximal contact, compromised chewing function due to tooth sensitivity, and vulnerability of teeth to incomplete eruption. Oral reconstruction of these patients requires a multidisciplinary approach, including periodontal and orthodontic procedures, orthognathic surgery, endodontic treatment, and restorative and prosthodontic rehabilitation.

Conclusion: A two-year follow-up observation confirmed satisfaction, appropriate function, and esthetics. **Keywords:** Amelogenesis imperfecta, Prosthetic reconstruction, Orthognathic surgery

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Introduction

The term "amelogenesis imperfecta" (AI) refers to a genetic disorder characterized by quantitative or qualitative enamel defects without systemic complications or metabolic disorders. ¹There are three main subgroups of AI: hypoplastic, hypocalcified, and hypomature.

As a result of hypoplastic AI, the affected teeth exhibit a rough, pitted surface with grooves and a reduced thickness of the enamel layer due to deficiencies in enamel construction. Hard and translucent enamel with some grooves is evident. Reduced hardness can be present in subjects with hypocalcified AI, but the enamel layer demonstrates ordinary thickness and is frequently yellowish-brownish in color. Opaque or chalky enamel, which wears away rapidly, is characteristic. The enamel layer related to hypomatured forms of AI illustrates standard thickness, but the enamel is slightly softer, mottled, and often chipped away (1-4).

Patients with AI are also affected by gingivitis, tooth sensitivity, deposition of calculus, anterior open bite, excessive wear and decrease of occlusal vertical dimensions, poor esthetics, loss of proximal contact, compromised chewing function due to tooth sensitivity, and vulnerability to incomplete eruption (5,6). Patients with AI are reported to have several psychosocial effects, such as higher anxiety and depression and lower selfesteem (7).

Treatment of AI is a multidisciplinary approach, and it includes periodontal consideration, orthodontic procedures, orthognathic surgery, endodontic treatment, crown lengthening, and restorative and prosthodontic treatment (2,8). In these patients, there is a high incidence of generalized gingivitis. Sensitive tooth structure is associated with poor oral hygiene and periodontal disease. Scaling and root planning are considered appropriate treatment options. Furthermore, crown lengthening is recommended when a patient has short teeth and gingival hyperplasia (2,7).

On the first visit, it is often discovered that patients have anterior open bite, different vertical growth patterns, and malocclusion (2,9). Orthodontic treatment is recommended in these cases, but enamel deficiency can reduce bonding, particularly in hypoplastic cases. Alternatively, orthognathic surgery may be employed,



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which may result in the extraction of some teeth (2,9).

Restorative and prosthetic care can resolve several associated complaints related to occlusal wear, interdental spaces, tooth discoloration, and aesthetic needs. Direct composite restorations are considered temporary treatment options, especially in the case of hypomature or hypomineralized teeth. In terms of treatment options, stainless steel crowns are recommended for children, while porcelain-fused-to-metal or all-ceramic crowns are recommended for adults (2,10).

Case Report

A 19-year-old woman was referred to the Department of Prosthodontics by a general dentist. Their chief complaint was pain caused by worn teeth on the upper left side and an unsatisfactory aesthetic. She had had discolored teeth since early childhood. An extraoral examination revealed a long face appearance with an increase in the height of the lower third of the face. The Class II appearance was evident from a profile view. The frontal retracted view revealed an anterior open bite (8 mm), no anterior guidance, posterior bilateral crossbite, uneven gingival margin, midline discrepancy, and misshapen, short clinical crowns (Figure 1). The occlusal view of the upper arch exhibited an omega-shaped and constrictive arch, which are common abnormalities in amelogenesis cases (11).

Clinical and radiographic examination of the patient revealed short clinical crowns; tooth #11 was impacted, and teeth #1, 16, 17, and 32 were semi-impacted. Moreover, root proximity between teeth #18 and 19 and #30 and 31 was evident. Carious lesions were found on teeth #13 and 14 (Figure 2). No abnormality was detected in temporomandibular joint movements. Papillary and marginal gingivitis were present, especially in the anterior mandible, and mouth breathing was suspected to be an etiologic factor.

The patient was then referred to the Oral and Maxillofacial Medicine Department with a diagnosis of hypomaturation and hypocalcification AI. Attrition was thought to be caused by enamel hypomaturation, particularly in the posterior teeth. Furthermore, a trial etching was performed to detect calcification in the tooth and plan the treatment. This test revealed an unfavorable bonding surface.

In lateral cephalometry, U1 to SN (116) was greater than normal (103), but SNA (82) was within the normal range (78–84) (12). Because of the difficulty in achieving a predictable bond area and the patient's rejection of orthodontic treatment, bimaxillary orthognathic procedures were planned after an orthognathicorthodontic consultation.

All impacted third molars and deciduous canines were extracted in the first step. A third molar extraction was necessary to facilitate the healing of the bone before bimaxillary orthosurgery. Next, the crown height of the teeth was evaluated, and crown lengthening (CL) was recommended for clinical crowns shorter than 3 mm for anterior teeth and premolars and crowns shorter than 4 mm for molars. These occlusocervical dimensions provide adequate resistance for crowns (13). After assessment, CL was performed on the mandibular and maxillary premolars, molars, and maxillary canines. The patient was then prepared for orthognathic surgery.

Before orthognathic surgery, some procedures should be performed to establish ideal occlusion and allow adequate space for restorative materials. The conservative preparation of the teeth, arbitrary mounting and waxingup, anterior and posterior temporization, and fabrication of surgical stents should all be considered.

In the first step, the tooth was prepared to eliminate undercuts. It was essential to conservatively prepare the CAD-CAM provisional restorations to reduce the risk of pulp sensitivity and ensure adequate strength during the healing process.

Anterior teeth contours and height were confirmed using composite veneers and were assessed considering the upper lip, aesthetics, and speech. The impressions (alginate, cromogel, Marlic, Iran) were then made and poured using type IV dental stone (Silky-Rock; Whip Mix Corp, Louisville, KY). Diagnostic casts were mounted on a semi-adjustable articulator (Whip Mix Corp, Louisville, Ky) using an ear-bow transfer (Whip Mix Corp) and a centric relation record (Kerr Corp, Orange, Calif). The posterior maxillary wax-up was completed according to the average size of the teeth for esthetic purposes and



Figure 1. Intraoral frontal view

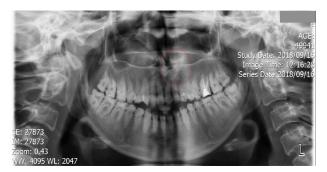


Figure 2. Initial panoramic view

adequate restorative thickness, and the mandibular waxup was completed following the upper wax-up based on width and height. Temporary PMMA crowns were prepared for posterior teeth using CAD-CAM technology (Figure 3). In the try-in session, impressions (alginate) were made for the temporary crowns, and casts were mounted in a semi-adjustable articulator (Whip Mix Corp, Louisville, Ky) using a face-bow transfer and interocclusal record. Cast surgery was performed to simulate surgical steps and fabricate surgical stents.

Before model surgery, all the reference lines were drawn. Then, the maxillary cast was mobilized and stabilized on the considered position, and the first occlusal splint was made. Afterward, the mandibular cast was mobilized, brought into occlusion with the maxillary cast, and stabilized, and a second surgical stent was prepared (Figure 4). In the present case, two surgical stents were made because it was a double-jaw surgery. The treatment plan included a segmented Le Fort I maxillary osteotomy with 5 mm posterior impaction and bilateral sagittal split osteotomies (BSSO) for 5 mm mandibular advancement, 1 mm midline shift, and counterclockwise rotation. Then, the patient underwent orthognathic surgery and rigid fixation. In orthognathic surgery cases, postoperative fixation of the segments with miniplates and screws and light elastics are used to guide jaw function after the operation (14,15). After the surgery, the patient used the provisional restorations for 6 months without any complications, but after that time, there was approximately a 1 mm relapse of the surgery in the vertical direction.

Prophylaxis and oral hygiene instructions were provided before treatment. Additionally, endodontic treatment was performed on teeth #13 and 14. Afterward, full-arch impressions of the maxillary and mandibular teeth were made using condensation silicon (Speedex; Coltene, Switzerland) and poured using type IV dental stone. Centric relation was recorded with a bimanual manipulation technique using an acrylic anterior deprogrammer (Pattern Resin LS, GC Dental Corp) and bite registration silicone (Futar D; Kettenbach GmbH &Co). The centric relation position was recorded using an interocclusal record, and the casts were mounted on a semi-adjustable articulator (Dentatus ARH-Type; Dentatus AB) by an arbitrary facebow (Dentatus Facebow) using an ear-bow transfer.

Accordingly, custom-cast posts and cores were fabricated on teeth #13 and 14. Under local anesthesia, the teeth were prepared with a circumferential radial shoulder margin configuration (Figure 5). Following this, PMMA provisional restorations were designed using the EXO CAD software program. Next, they were printed and cemented with temporary cement (Temp Bond, Kerr Corp., Orange, CA, USA). As a result of evaluating provisional restorations and establishing anterior guidance (Figure 6), definitive impressions of the prepared maxillary and mandibular teeth were obtained using a two-phase impression technique, putty, and extra-light body impression materials (Panasil,



Figure 3. Provisional restorations before orthognathic surgery



Figure 4. Model surgery



Figure 5. Occlusal view of final tooth preparation



Figure 6. Provisional restorations after orthognathic surgery

Kettenbach GmbH & Co). The casts were mounted using the cross-mount technique. The temporary crowns were subsequently replaced by zirconia restorations (Katana Zirconia HT, Kuraray Noritake Dental Inc., Tokyo, Japan) shade A1, which were fabricated by CAD-CAM technology and layered by porcelain (CERABIEN[™] ZR, Kuraray Noritake Dental Inc., Tokyo, Japan). In the try-in session, the marginal fit and occlusion of the crowns were evaluated intraorally, and a canine-protected occlusion was provided to the patient.

In the delivery session, the crowns were cemented with temporary cement (Temp Bond, Kerr Corp., Orange, CA, USA) (Figure 7). Impressions of two arches were taken by alginate, and in the next session, a dual cure acrylic night guard was made to protect teeth and restorations against possible bruxism. The occlusal adjustment required three-month recall evaluations for one year. After confirmation of occlusion, all crowns were cemented with glass ionomer cement (Fuji II, GC Dental Corp). At the two-year follow-up, the patient was satisfied with the aesthetics and function, and the gingival margins were stable with no inflammation or recession (Figure 8).

Discussion

A multidisciplinary approach is required to treat AI, and a variety of treatment options should be considered, including:

- 1. Conservative restorative treatment such as direct composite or SS crown for children
- 2. Orthodontic treatment
- 3. Orthognathic surgery
- 4. Definitive restorative treatment and indirect restorations (2,9,10)

A combination of orthognathic and periodontal surgery was performed in the present case before prosthodontic reconstruction. Due to hypocalcification of the teeth, bracket bonding could not be successful in orthodontic treatment. The present case involved orthognathic surgery, including a counterclockwise rotation and advancement of the mandible and posterior impaction of the maxilla.

As part of this study, CAD-CAM provisional restorations were used during the healing phase of

orthognathic surgery. These restorations stabilized occlusal contacts and reduced the risk of gingival tissue growth, CL relapse, and teeth extrusion. The durability and mechanical properties of provisional PMMA CAD-CAM restorations are important characteristics. Furthermore, these restorations show improved hardness, surface roughness, flexural strength, and impact strength compared to conventional heat-cured types (16).

Relapse is one of the late postoperative complications associated with orthognathic surgery. Proffit mentioned that changes > 2 mm and > 2° toward the baseline values are considered clinical relapse, a multifactorial problem (17). Several factors influence the outcome of surgery, including the presurgical diagnosis, the surgeon's skill, the correct surgical technique, the type of fixation, and the patient's individual features, including muscle and soft tissue tension and the magnitude of mandibular movement (18-20).

According to Fonseca, the most important risk factors for immediate relapse are TMJ problems, condylar displacement, and a failure to reposition the disc before orthognathic surgery. Even though mandibular advancement is a stable orthognathic procedure in patients with short or normal face heights, condylar resorption after mandibular advancement results in a relapse into an anterior open bite (21); additionally, researchers agree that in a bimaxillary procedure, there is a greater degree of relapse in the mandible than in the maxilla (22). Long-term relapses have also been associated with a large number of mandibular advancements beyond neuromuscular adaptation and counterclockwise movement (22). In the present case, counterclockwise movement may have been the cause of the relapse. An important point to note is that light elastic therapy after orthognathic surgery is indicated in cases of bimaxillary surgery to minimize molar extrusion, anterior open bite, and CO-CR discrepancy (15).

The use of porcelain-fused-to-metal or full-ceramic restorations is considered an acceptable prosthetic option in the reconstruction of AI cases. According to several authors, full porcelain restorations are the best option in terms of aesthetics and periodontal preservation (2). Zirconia restoration is considered a strong and durable



Figure 7. Frontal view of final restorations



Figure 8. Final panoramic view

framework that presents high flexural strength exceeding 1200 MPa, excellent biocompatibility, and low wear characteristics compared to sound teeth (23,24), and for these reasons, zirconia crowns were selected in the present case.

A cross-sectional study comparing the survival rate of restorations in AI patients and control groups found an 80% survival rate in the unaffected group after five years, while the survival rate in patients with AI was only 50%. As a result, the replacement rate of restorations in AI patients was approximately 2.5 times higher than in unaffected patients. Additionally, regarding the longevity of restorations, the survival rates for patients with hypoplastic types of AI appear to be higher than those with hypomatured or hypocalcified dentition (25).

Conclusion

This article described the full-mouth rehabilitation of a patient suffering from AI. Anterior open bite, posterior bilateral crossbite, and short and malformed clinical crowns were some of the challenges in the present case. CAD-CAM provisional restorations after Orthognathic surgery demonstrated durability during the healing phase. Follow-up observation confirmed satisfaction, appropriate function, and esthetics.

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Authors' Contribution

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Visualization: Negin Yaghoobi.

Writing-original draft: Farideh Geramipanah, Negin Yaghoobi.

Competing Interests

The authors declare that they have no conflict of interest.

Ethical Approval

Consent for publishing photographs was acquired from the patient.

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