

Full-mouth Rehabilitation of an Amelogenesis Imperfecta Case with Skeletal Open Bite

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Submitted: 2019-7-02; **Accepted:** 2019-09-17; **Published Online:** 2019-10-04; **DOI:** 10.22037/rrr.v4i4.29675

Introduction: Amelogenesis imperfect (AI) refers to a group of autosomal dominant disorders primarily affecting the enamel and can present a variety of clinical appearances. This report describes a multidisciplinary approach for oral rehabilitation of a young man patient with hypoplastic AI with severe skeletal anterior open bite (AOB). **Case Report:** A 22-year-old male patient was presented with compromised masticatory efficiency and dissatisfaction with his teeth appearance. Based on the clinical and radiographic examination, the patient was confirmed as AI (hypoplastic type) with severe AOB. The treatment included orthognathic surgery (Le Fort I osteotomy and genioplasty) for correcting AOB, long face, and limited space in the posterior region, followed by crown lengthening and prosthetic rehabilitation. Before surgery, a mock-up of all teeth was performed to facilitate alignment and stabilization of the dental arches during the Le Fort I osteotomy. **Results:** Solid tooth interdigitation that provided with mock-up, helped to adjust the stable skeletal position of the maxilla during the surgery. **Conclusion:** preoperative mock-up appears to be an efficient fixation option to achieve an acceptable occlusion after prosthetic rehabilitation in AI patients with severe AOB.

Keywords: Amelogenesis Imperfecta; Open Bite, Orthognathic Surgery, Dental Prosthesis

Introduction

Amelogenesis imperfecta is a group of developmental conditions with genomic in origin, that primarily affects the quality and/or the quantity of the enamel structure and appears without any associated generalized or systemic anomalies (1). Different inheritance patterns for this abnormality have been reported including autosomal-dominant, autosomal recessive or X-linked mode of inheritance (2). The prevalence of AI has been reported to vary between approximately 1:700 to 1:16,000, depending on various diagnostic and demographic criteria used (3). Based on the most commonly used classification, AI can be categorized into four broad groups of hypoplastic, hypomaturation, hypocalcification, and hypomaturation hypoplasia with taurodontism (4).

The most challenging dental issue in adult AI patients is skeletal AOB. Several authors have reported that AOB commonly occurs in patients with AI (in both the hypoplastic and hypomineralization types) and cause insufficient posterior restorative space with minimal to no loss of occlusal vertical dimension (OVD) while there is an excessive restorative space in anterior region (5).

The prevalence of AOB among individuals with AI varies from 24% to 60% (6). Skeletal features and clinical characteristic of AOB in patients with AI are similar to the AOB subjects not suffering from AI. Omega-shaped maxillary dental arch, reversed mandibular curve of Spee, more constriction of dental arches, and transverse discrepancy between maxillary and mandibular arches has been seen in AI cases. All of these indicate that AOB in patients with AI is caused by a genetic abnormality in craniofacial development, rather than by local factors affecting alveolar growth (7).

There are various methods for severe AOB correction, including segmental orthognathic surgery, molar intrusion with skeletal anchorage, corticotomy-facilitated orthodontic treatment (8). Orthodontic and surgical correction of AOB is often difficult due to the high relapse rate, especially if associated with AI and can be one of the challenging and complicated therapeutic approaches in orthognathic surgery (9). Dental arch alignment in these patients is usually very difficult and sometimes causes special problems (10). The following clinical report presents step by step a modified surgical approach in treating a hypoplastic AI young male with skeletal AOB that enables handling and fixation of dental arches during surgery.

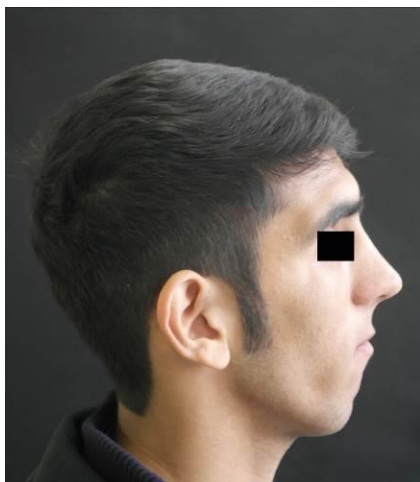


Figure 1. Preoperative lateral profile view of the patient with AI and Skeletal class II

Case Report

A 22-year-old male patient presented to the Prosthodontics Department with a chief complaint of discolored teeth and poor masticatory efficiency. The family history revealed that his mother, sister and brother had a similar problem, indicating the genetic origins of the condition. The medical history revealed no abnormalities. Extraoral examinations revealed long oval facial form in frontal view and a convex profile in lateral view, slight asymmetry and deficiency in chin, increased lower anterior facial height, and incompetent lips with a nasolabial angle of 110° (Figure 1). Maximum mouth opening measured 48 mm. Smile analysis revealed revers smile line, AOB, gummy smile and slight lower midline discrepancy (comparing to midline of the face and upper teeth). No remarkable findings were identified in temporomandibular Joint.

Teeth showed yellow-brown discoloration, pitted and irregular surface, excessive attrition, a short and tapered crown form, wide occlusal surfaces, spacing in the posterior regions but tight proximal contacts in the anterior region, and hypersensitivity either to cold or hot predisposes (Figure 2). Skeletal and dento-alveolar diagnoses included high palatal vault, omega-shaped maxillary dental arch, constricted maxillary dental arch especially in inter-premolar region, reversed mandibular curve of Spee. In vertical plan, patient had long face and AOB. In sagittal plane, because of midline deviation, patient had full cusp class II malocclusion in the right side and half cusp class II in the left side. In transverse plane, there was lingual cross bite from canine to first molar



Figure 2. Preoperative intraoral view. A) Mandibular occlusal view; B) Maxillary occlusal view; C) Frontal view

in left side because of maxillary constriction. On the right side the patient had no cross bite.

Periodontal examination revealed poor oral hygiene, calculus deposition on the lingual surface of the mandibular anterior teeth, moderate generalized gingivitis and localized periodontitis. The patient also had mouth breathing and tongue interpositioning habit with edematous and uneven gingival margin.

Panoramic radiograph view demonstrated wide pulp chamber and short root trunk, furcation proximity to the alveolar crest, the proximity of the roots while having a normal length, reduced enamel thickness with radiodensity that was approximately the same as that of dentin. All permanent teeth were present except right mandibular first molar. Clinical and radiographic examination confirmed him as a case of hypoplastic-type AI.



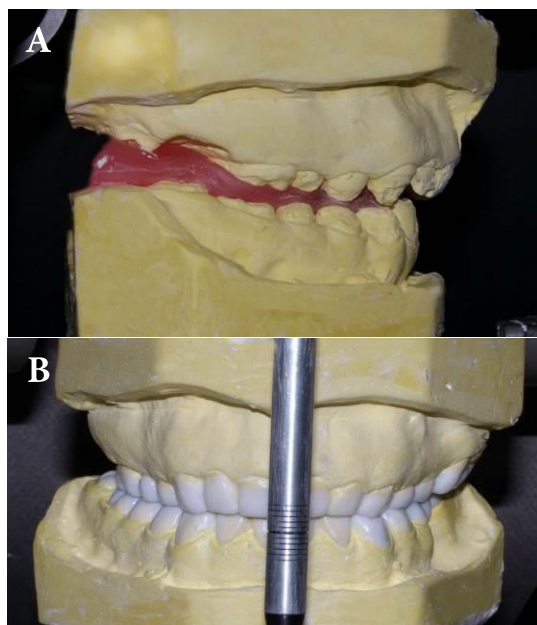


Figure 3. A) Arbitrary mounting; B) wax-up of all teeth based on the aesthetic and occlusal criteria

Pre-surgical treatment

Scaling and root planning was performed to treat the existing chronic gingivitis and periodontitis. Then oral hygiene instructions were also given to the patient. Caries was restored with glass ionomer (GC Corporation, Tokyo, Japan). The upper right second molar also was extracted.

Orthodontic treatment options were not possible because of less etchable of enamel surfaces and the conical form of the teeth resulting in difficulties in bonding orthodontic brackets. The diagnostic composite laminate veneer of the upper anterior tooth and crown-root ratio analysis showed that the AOB could not be corrected solely by restorative approaches. Therefore, after consulting with oral and maxillofacial surgeons, it was concluded that a Le-Fort I surgical procedure is needed to correct the AOB and limited posterior restorative space.

In the presurgical phase, maxilla and mandible casts were made and without bite registration and only based on the aesthetic criteria mounted on a Dentatus articulator (Dentatus USA Ltd., NY, USA). Wax-up of all dentition was performed (Figure 3), followed by impression of wax-up casts for construction of clear template which used for composite mock-up. The main purpose of this mock-up was to correct vertical height of teeth and create an occlusal intercuspation as a guide during the surgery for proper intraoperative jaw positioning while protecting the dentin/pulp complex (Figure 4).

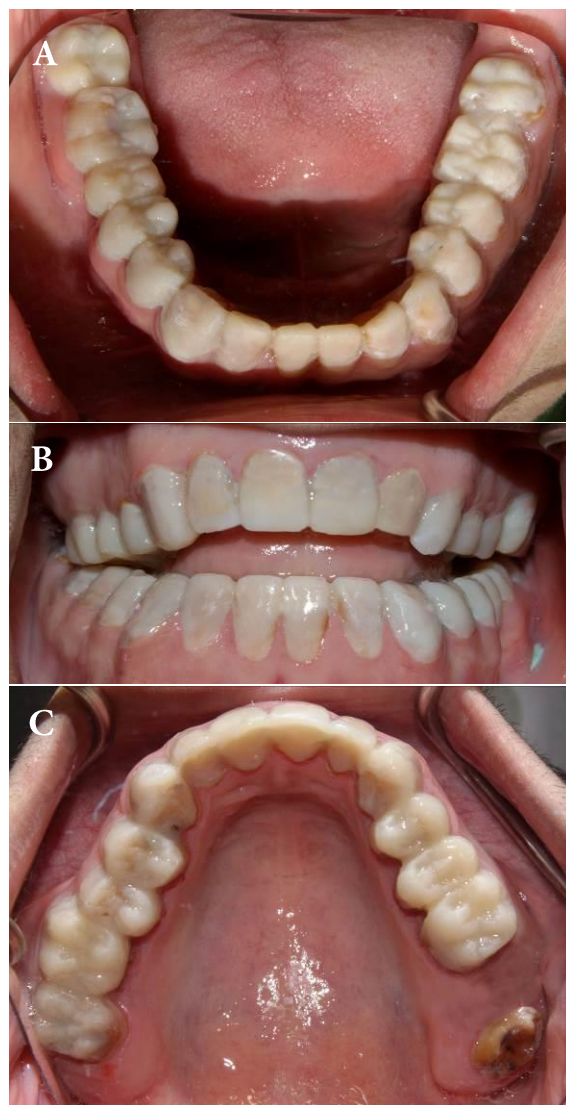


Figure 4. A) Mock-up of all teeth before orthognathic surgery; B) Mandibular occlusal view; C) Maxillary occlusal view Frontal view

After mock-up, to ensure of ideal and stable occlusal relationship, maxillary and mandibular impressions were made and casts were mounted on a Dentatus articulator (Dentatus USA Ltd, NY, USA) using a face bow and a model surgery was performed on the articulator (Figure 5).

Surgery

In order to correct AOB and create space in posterior region, one piece Le Fort I osteotomy was performed, posterior maxillary segments were moved 5mm superiorly, anterior maxillary segments were advanced 6mm and moved 1 mm inferiorly, and maxilla were rotated 1mm to the right side. Simultaneously, genioplasty was performed to reduce height of





Figure 5. Model surgery

the lower third of the face. The rigid internal fixation was then carried out using mini-plates (Figure 6). During the settling period, composite restorations were polished and modified. The patient was given oral health education. Figure 7 demonstrates post-operative improved facial harmony and intraoral view after surgery and polishing.

After ten months of consolidation period, impression of each arch was made. The surgical pattern was constructed according to the patient's smile line, esthetic analysis, and furcation involvement and then used for crown lengthening surgery and leveling of the gingival margin.

During crown lengthening, the root of the mandibular left first molar was exposed so autogenous bone grafting was simultaneously performed on the defect. It was also decided to extract mandibular left third molar due to its inappropriate crown-root ratio and lack of the attached gingiva on the distal side of this teeth.

Prosthetic phase

Two month later, a primary impression was made. Study casts was poured and mounted on a Denar Mark II articulator (Whip Mix, Kentucky, USA) using a face bow. Posterior determinant of the occlusion was recorded using the lateral records. A diagnostic wax-up of canines and incisors was made, and the occlusal plane was established by the broadrick flag. According to the occlusal index, the wax-up was completed.

Under local anesthesia, all teeth were prepared (with heavy chamfer finish line) for receiving the all-ceramic

prosthetics. Preparation depth was controlled with putty index of this wax-up. Due to the extensive destruction of the tooth structure, post patterns were fabricated for the upper left first and second molars, and the right third molar. The casting posts were cemented with glass-ionomer cement (GC Corporation, Tokyo, Japan).

All teeth were temporary restored with acrylic provisional restorations. The patient's occlusion with temporary crowns was mutually protected occlusion. Casts of the temporary crowns were mounted on the Denar Mark II articulator and mechanical anterior guidance table was set. Movement settings were transferred to the Cercon computer-aided design/computer-aided manufacturing (CAD-CAM) system software (Dentsply, sirona prosthetics, Pennsylvania, USA). Final maxillary and mandibular full arch impressions were made with an additional silicone impression material (Panasil, Kettenbach, Eschenburg, Germany) along with cross-recording.

The master casts were poured and every-other mounting were performed in the dental laboratory. The temporary casts were scanned and mounted on the virtual articulator in the software. Subsequently, master casts also were scanned and cross-mounting were performed virtually. Full anatomic wax-up and cut back was performed on the virtual articulator. Then zirconia frameworks (Zirkonzahn, Australia) were fabricated.

Frameworks were tried in and by placing frameworks on the tooth alternatively, periapical radiographs were taken to ensure correct seating of them. An incisal edge index and a customized anterior guidance table of temporary casts were made in Denar articulator and base on porcelain (Noritake Feldspathic porcelain, A2) was added to the frameworks.

The restorations were evaluated for fit, esthetics, phonetics, and centric and eccentric occlusal contacts. The patient's reverse smile line was corrected (Figure 8). After glazing, the restorations were cemented with temporary non-eugenol cement (Kerr, Orange, CA, USA).

Oral hygiene instruction was given to the patient and was recall 24 hours later. He reported no complications and no complaints. One month later, permanent cementation was performed with glass ionomer cement (GC Corporation, Tokyo, Japan) (Figure 9). The patient was recommended to recall evaluations at 3-month intervals for 12 months and then at one-year intervals. He was very satisfied with the treatment results.



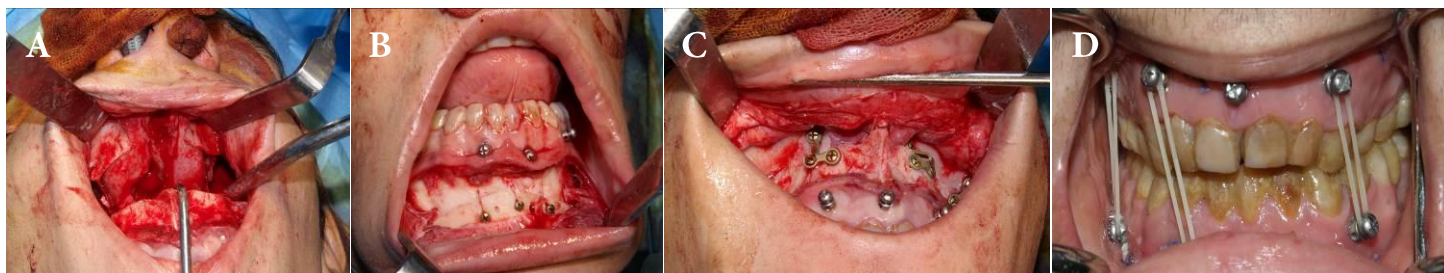


Figure 6. A) Lefort 1; B) Genioplasty; C) Rigid fixation with miniplate; D) One month after surgery

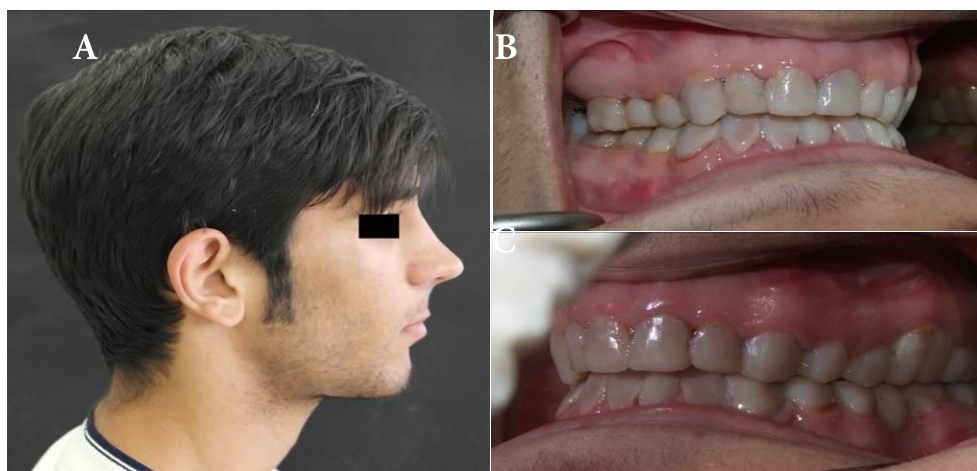


Figure 7. A) Postoperative lateral profile view, intraoral view after surgery, B) Left; C) Right

Discussion

In patients with mild to moderate AI, a simple and effective treatment option is the use of the natural stratification of composite resins to mask deficient enamel and mimic the tooth's natural appearance (10). However, presented cases showed less etchable of enamel surfaces with vertical discrepancy that caused composite veneer and ceramic laminate alone would not accomplish this goal.

Orthodontic treatment in patients with severe AOB can be associated with either orthognathic surgery or skeletal anchorage system (dental implants, screws, and miniplates) (11). The molar intrusion with skeletal anchor increases overbite without elongation of anterior teeth, and decreases the height of the lower face by rotating counterclockwise of the mandible. Kuroda et al. suggested that treatment of severe AOB cases by molar intrusion with skeletal anchorage was easier and more useful than that with orthognathic surgery (11).

Corticotomy and compression osteogenesis in the posterior maxilla was recently established as a trustworthy alternative for

treatment of severe AOB (12). Another name of this technique is corticotomy-facilitated orthodontic treatment using skeletal anchorage. This technique utilizes a gradual postoperative compressive force to adjust the ideal position of the bone/teeth segments over a shortened treatment period (13). Orthodontic treatment is not possible in the present case because of the condition of the enamel and lack of crown height; as a result, the latter two techniques are not applicable.

During the past decades, Le Fort intrusion osteotomy with or without genioplasty or bilateral mandibular sagittal split osteotomy is considered a relatively routine intervention for AI patients with severe AOB (14). Therefore, we decided to combine orthognathic surgery and prosthetic rehabilitation for treatment of this patient. The purpose of surgical treatment of adult AI patients with AOB is to provide appropriate intermaxillary relationship to facilitate occlusal rehabilitation. Restorations are usually needed to restore the crown anatomy and reduce the risk of pulp damage (15).

Alignment of the dental arches and maintaining of the segments in a desired position during surgery makes special problems in AI patients due to the reduction of the thickness





Figure 8. Smile view A) Before; B) After treatment



Figure 9. A) Post treatment of intraoral view; B) Left lateral view; C) frontal view (Right lateral view)

and hardness of the enamel and conical shape of teeth and difficulty of applying the ligatures around the abraded short (pre-) molars (16). If the height of the crowns allows, a full cover occlusal splint can be used to maintain the segments in the proper position. The reduced crowns height in AI patients, like this patient, makes it difficult to use occlusal splint (15). On the other hand, it is very important for the surgeon to inform about the prosthetic treatment plan. The surgeon determines the vertical position of the maxilla based on the relationship between the upper lip and the incisal edge of the upper incisors, taking into account the crowns that might be reconstructed in the future. Also, because of the abraded short crowns in these patients, it would often require that the surgeon create a larger than normal freeway space (17).

In the present patient, mock-up was performed preoperatively to facilitate alignment and fixation of the dental arches during the surgery by creating a stable occlusion. It is expected mutual contacts between anterior teeth and the solid interdigitation in (pre-) molar regions that is the best guarantee for post-surgical stability, to be provided with mock-up. As well as, with this technique, the surgeon was able to inform about the planned prosthetic rehabilitation during the surgery.

The restorative management of AI depends on the patient's age, socioeconomic status, type and severity of the disorder and the intraoral situation (18). Full coverage crown is usually advised as a definitive restoration in the most severe hypocalcified AI cases (19). Even if a significant loss of tooth structure is required; these restorations protect the tooth from further damage of the poorly mineralized and friable enamel. In the treatment of this patient, full coverage zirconia restoration for inherited esthetics, excellent biocompatibility and improved physical properties was selected (20).

Conclusion

AI is commonly associated with skeletal AOB which is the most complicated dental problem. Treatment of AI patient with severe skeletal AOB is mainly associated with surgically repositioning the maxilla and/or the mandible. In the present case, composite mock-up performed before surgery. Our results indicate a preoperative mock-up can facilitate the alignment and maintaining of the dental arches in a desired position during the orthognathic surgery to correct AOB.

Conflict of Interest: 'None declared'.



Regeneration, Reconstruction & Restoration 2019;4(4): 170-176



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Please cite this paper as: Nazari V, Alikhasi M. Full-mouth Rehabilitation of an Amelogenesis Imperfecta Case with Skeletal Open Bite. *Regen Reconstr Restor.* 2019;4(4): 170-176. Doi: 10.22037/rrr.v4i4.29675.

