# A Multidisciplinary Approach to the Functional and Esthetic Rehabilitation of Amelogenesis Imperfecta and Open Bite Deformity: A Case Report

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#### ABSTRACT

The treatment of amelogenesis imperfecta (AI) with an anterior open bite (AOB) is a challenge for the clinician and often requires a multidisciplinary team of specialists. Most often, patients suffering from these conditions are young and a good functional and esthetic long-term result must be aspired. This clinical report illustrates the orthodontic, maxillofacial, restorative, and prosthodontic rehabilitation of a 20-year-old woman with a hypoplastic form of AI and an AOB malocclusion, having received treatment for the last 6 years. It included adhesive resin composite restorations, orthodontical and maxillofacial surgery with a one-piece Le Fort I osteotomy, and a genioplasty. Subsequent prosthodontic therapy consisted of 28 all-ceramic crowns whereby a solid interdigitation, a canine guidance, and consistent and regular contacts between tooth crowns could be achieved to assure a good functional and esthetic oral situation. The tooth preparation techniques guaranteed minimally invasive treatment. The patient was affected very positively.

#### CLINICAL SIGNIFICANCE

This article describes an interdisciplinary approach to the successful treatment of a patient with a hypoplastic form of amelogenesis imperfecta over a period of 6 years. It starts with a discussion of the conservative steps taken during adolescence and concludes with the final prosthetic rehabilitation with all-ceramic crowns after reaching adulthood.

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INTRODUCTION

renetically determined and rare Jdysplasia of the enamel formation are known as amelogenesis imperfecta (AI) and have been classified into several groups by various authors.<sup>1-4</sup> The most widely accepted classification contains four groups and is based on the kind of development dysfunction of the enamel formation:<sup>2</sup> class I) disturbance of the translation and secretion of an extracellular matrix (hypoplasia), class II) disturbance of the maturation of enamel (hypomaturation), class III) disturbance of the matrix mineralization (hypocalcification), and class IV) hypomaturation-hypoplasia in combination with taurodontism.

AI interferes with normal enamel formation in the absence of a systemic disorder and generally affects all or nearly all teeth in both the primary and permanent dentitions. Dentin formation is not affected. A hypoplastic AI shows a reduced enamel thickness with normal hardness. Where present, enamel radio-opacity is normal. A hypomature or hypocalcified AI has a normal enamel thickness which is softer, especially for the hypocalcified form. The lower mineral content leads to reduced radio-opacity. Generally, teeth may be discolored, sensitive, brittle, and showing atypical crown morphology.

AI may be inherited by autosomal dominant, autosomal recessive, x-linked, or sporadic inheritance patterns.<sup>5</sup> Diagnosis involves the exclusion of extrinsic environmental or other factors, establishment of a likely inheritance pattern, recognition of phenotype, and correlation with the dates of tooth formation to exclude a chronological developmental disturbance.6 The estimated prevalence of AI depends on diagnostic criteria as well as population, and is estimated to be between 1:700 and 1:14,000.7-10

Even though AI is by definition a disorder of enamel, it has been associated with several other dental anomalies including disturbances in eruption, congenitally missing teeth, anterior open bite (AOB), pulpal calcifications, pathologic root and crown resorption, and taurodontism.11 While the incidence of AOB is commonly attributed to skeletal and soft tissue anomalies,<sup>12</sup> embryological investigations suggest that enamel and the craniofacial skeleton may share a common ectomesenchymal origin.<sup>13,14</sup> The incidence of AOB in patients with AI varies from 24 to 60%.9,15 Witkop and Sauk9 suggested that tongue interposition, probably provoked by an increased sensibility of the teeth, results in an impeded alveolar growth by the tongue interpositioning. Rowley and colleagues,<sup>15</sup> however,

proposed that the frequent association of AOB and AI is caused by a genetically determined anomaly of craniofacial development rather than by local, mechanical factors influencing alveolar growth.

A complex rehabilitation of a patient with AI and an AOB needs a multidisciplinary team of a pediatric dentist, an orthodontist, a maxillofacial surgeon, and a prosthodontist. In the primary and mixed dentitions, the team must aim at reducing sensibility and preventing attrition of the teeth as well as at ensuring the patient's oral health and prophylaxis. In the permanent dentition, advanced restorative techniques, often in combination with orthodontic treatment and maxillofacial surgery, must be used to restore and protect esthetic and functional aspects for the long-term situation, ranging from a stable chewing function to facial harmony.<sup>16-18</sup> During the generally long-term treatment and care of the patient (and parents), the prosthodontist should play a key role in the multidisciplinary team because his involvement includes all aspects of the case planning and treatment.<sup>19</sup>

#### CASE REPORT

## **Case Presentation**

An otherwise healthy patient was diagnosed with hereditary AI and an AOB. She was referred to the dental clinic of the University of



Figure 1. Initial facial photograph at the age of 14.

Bern at the age of 14 by her pediatric dentist. Her father and both her brothers are suffering from AI with an AOB, too.

## Extraoral and

### **Physiognomic Aspects**

The patient showed a straight profile with a pronounced chin and jaw, a strong gummy smile of 5 to 6 mm, an increase of the dimension of the lower face, and no bite collapse (Figure 1). The temporomandibular joint displayed an anterior discus displacement with repositioning. Because the patient reported no discomfort or pain, this situation needed no treatment.

#### Intraoral Aspects

A complete dentition was present with a generalized AI, classified as a hypoplastic form with an almost complete aplasia of enamel (Type I-G after Witkop<sup>2</sup>) (Figure 2a–e). In a panoramic radiography, the thin enamel layer could not be distinguished from the underlying dentin (Figure 3). Tooth crowns exhibited a distinct microform with no interdental contacts (large spacings) whereas the roots showed normal length and form. The pulp chambers were regular in size. Neither fillings nor caries existed. In the vertical dimension, the bite was open up to and including the second premolars on both sides with an overbite of -3 mm. Skeletal growth was normo-divergent with an ML/NSL angle of 35.5°. In the transversal dimension, the first and second molars were in an edge-to-edge bite, and the middle line in the lower jaw was displaced to the left by 2 mm. In the sagittal dimension, a skeletal class I (ANB 2°, Wits-appraisal -3 mm) was present with the molars showing a neutral occlusion on the right side and a distal one on the left side (Figure 2d and e). The patient had a habitual tongue interposition and suffered from dentin hypersensibility. Her oral hygiene was good with no signs of gingivitis.

The initial restorative treatment was decided to be conservative, using direct adhesive materials (MIRIS, Coltène Whaledent, Altstätten, Switzerland) to restore the anatomy of the crowns by replacing the missing enamel and thereby reducing dentin sensibility (Figure 4a–c). This treatment



Figure 2. Intraoral views before treatment: A, frontal view, B, occlusal view of the maxilla, C, occlusal view of the mandibula, D, right side view, and E, left side view.



Figure 3. Panoramic radiography before treatment.

proved to be extremely difficult because of the conical crown anatomy and the aforementioned large spacings between individual teeths. Eighteen months later, a reevaluation showed that the AOB situation had worsened because of skeletal growth reasons (hyperdivergent with an ML/NSL angle of 38.5°) as well as because of the conservative treatment, and the bite had opened by 2 more millimeters to -5 mm. Therefore, a planned orthodontic treatment was postponed until the patient completed adolescence and a combined orthodontic and maxillofacial therapy would be possible to close the AOB. During the waiting period, the patient developed a slight gingivitis and small interdental carious lesions despite a good oral hygiene and an intensive recall plan. Both problems were treated; they originated because interdental hygiene had become difficult after the initial restorative treatment with unavoidable interdental and buccal overcontours.

# Orthodontic and Maxillofacial Procedure

After having reached completion of bone growth at the age of 18, an orthodontic reevaluation took place. In the vertical dimension, the open bite up to the first molar was unchanged, and the skeletal growth hyperdivergent with the ML/NSL angle at 38.5°. In the sagittal dimension, there was a skeletal class I (ANB 1.5°, Wits-appraisal



Figure 4. A, Anterior view and occlusal views of B, the maxilla and C, the mandibula after initial restorative therapy with adhesive resin composite.



Figure 5. Panoramic radiography after maxillofacial surgery with a one-piece Le Fort I osteotomy and a genioplasty.

-2 mm) and a dental class II on the left side. The transversal dimension showed an edge-to-edge bite between the molars on the left side and a displacement of the middle line in the lower jaw by 2 mm to the left. For the correction of the transversal relation, either a forced expansion or a three-piece Le-Fort I osteotomy was discussed. Because of the completion of bone growth, forced expansion was not an option. Because of a needed correction of only 3 mm, a surgical three-piece Le-Fort I procedure needed reevaluation after orthodontic treatment, with an

alternative option of a one-piece Le-Fort I osteotomy.

The patient and her parents agreed on a proposed orthodontic and maxillofacial treatment plan to close the AOB. It included four steps of (1) an orthodontic correction of tooth dislocations; (2) a three-piece osteotomy in the upper jaw combined with a genioplasty to adjust sagittal, vertical, and transversal dimensions; (3) a postsurgical orthodontic refinement; and (4) a prosthodontic rehabilitation to stabilize the achieved results.

After the presurgical orthodontic treatment, the transversal relation could be dentally corrected and only a one-piece Le Fort I osteotomy was required. After downfracturing of the maxilla, the appropriate amount of bone was removed at the lateral and medial sinus wall as well as the nasal septum to allow for the planned intrusion of the maxilla of 5 to 6 mm in the molar region and 2 to 3 mm in the incisive region.<sup>20,21</sup> The ostoesynthetic fixation was realized using MODUS mini-plates (Medartis AG, Basel, Switzerland). The genioplasty included an osteotomy with a caudodorsal positioning of the chin and again osteosynthetic fixation with MODUS mini-plates (Figure 5). To guide the occlusion, elastics were used for 2 weeks.

With the said maxillofacial surgery, the AOB could be closed completely, the gummy smile was reduced substantially, and a good facial harmony was established. Five months later, debanding and



Figure 6. Side views A, right and B, left after orthodontic and maxillofacial treatment with spacings between individual teeth.



Figure 8. Mock-up to determine esthetic parameters.

Figure 7. Diagnostic wax-up.

debonding took place, and deepdrawing templates in both jaws were incorporated to stabilize the achieved orthodontic and maxillofacial results. After these steps, a nonsolid interdigitation—because of the flat occlusional relief and tooth spacings—remained to be corrected by prosthodontic procedures (Figure 6a and b).

#### **Prosthodontic Procedure**

For diagnostic reasons, a wax-up was produced with a 1-mm vertical increase of occlusion (Figure 7), which was important to create enough space for a restorative reconstruction. However, the existing vertical dimension without a reconstruction was at its upper limit already and could not be increased arbitrarily. With a mockup, esthetic parameters could be determined intraorally (Figure 8).

The upper left canine needed a gingivectomy to harmonize the smile, and the four infraocclusioned and AI-affected wisdom teeth were extracted (Figure 9). Because of their advantages concerning esthetics, lesion-related preparation techniques, and the young patient's large pulp chambers, all-ceramic lithium disilicate crowns (e.max, Ivoclar, Schaan, Liechtenstein) were used in combination with adhesive bonding.

Under local anesthetics, all teeth were prepared with chamfer margins of 0.5 to 0.7 mm circumferentially and an occlusal reduction of 1.5 mm. Preparation depth was controlled with thermopressed sheets taken from the wax-up. The provisional restorations were prepared by the dental technician. They were fitted intraorally with the self-curing methylmethacrylate resin GC



Figure 9. Extracted wisdom tooth clearly showing the extent of amelogenesis imperfecta.

Unifast TRAD (GC America, Alsip, IL, USA) and fixed with the temporary, transparent cement TempBond Clear (Kerr, Orange, CA, USA) (Figure 10a–d).

After 2 months, the provisional restoration showed no signs of occlusional or bite relation changes, the temporomandibular joint was pain free, and the final restoration could be started, first in the upper jaw and 3 months later in the lower jaw. After insertion of thin retraction cords (000, Ultrapack, Ultradent, South Jordan, UT, USA) around all teeth, final impression was taken with an elastomer material (Impregum, 3M Espe, Seefeld, Germany). Occlusal registration was obtained with a hard addition-type A-silicon material (Futar D Occlusion, 3M Espe) with a slightly reduced vertical dimension from the provisional restoration. The working casts were mounted on an articulator (Kavo Protar Evo, Kavo, Biberach, Germany), and the framework for each tooth was waxed and pressed individually in an Austromat D4 oven (Dekema, Freilassing, Germany) (Figure 11a and b). Onto the pressed all-ceramic frameworks, two dentin firings and one final firing were applied (Figure 12). The ceramic crowns and the teeth were etched, primed, and bonded according to manufacturers' instructions before being adhesively fixed with a dual-cure polymerization resin cement

(VarioLink II, Vivadent, Schaan, Liechtenstein). The occlusion could be grinded using the provisional restoration in the mandible.

The restoration in the mandible with all-ceramic crowns was done using equal procedures and materials as for the maxilla. To guarantee the retention of the orthodontic and maxillofacial treatment, a retainer was palatinally/lingually fixed to the front teeth from canine to canine in both jaws.

With the prosthodontic rehabilitation, a solid interdigitation, a canine guidance, and consistent and regular contacts between tooth crowns could be achieved (Figures 13a–e and 14). The patient was very happy with the esthetic and functional result (Figure 15).

At the 1-year recall, the situation was esthetically, clinically, and radiologically unchanged, and no pathology associated with the rehabilitation was found.

#### DISCUSSION

The extensive rehabilitation of a young patient with a generalized AI in combination with an AOB is a challenge for any clinician, and a multidisciplinary team of dentists needs to be involved in the care plan. Several factors have to be taken into consideration, including the often young age of the patient, the quality and



Figure 10. The provisional restorations fitted intraorally, A, viewed from the front and B, from the side, as well as prepared by the dental technician for the C, maxilla and D, mandibula.



Figure 11. Framework for each tooth, waxed and pressed individually for A, the maxilla and B, the mandibula.

quantity of existing enamel and tooth substance, the periodontal condition, the long-term prognosis and stability of the result, and the total cost of treatment. The multiple treatment phases often last several years, and at each stage, the long-term consequences, risks, and benefits of the various therapy options must be discussed with patients and parents.<sup>11</sup>



Figure 12. All-ceramic lithium disilicate crowns.

#### **Open Bite Deformity**

The prevalence of open bite malocclusions in people with AI is greater than in the general population.<sup>22</sup> AOB with AI seems to be of skeletal origin<sup>14</sup> and not associated with deviations of dento-alveolar growth<sup>9</sup> or with failures in the eruption mechanism.<sup>1</sup> Patients with AOB and AI behave like patients with AOB who do not suffer from AI; skeletal and soft tissue components in both groups are comparable, but treatment of AOB in patients with AI is even more complicated and challenging because presurgical orthodontics are often not feasible because of lack of crown height and the condition of the enamel. A one-piece or multisegment Le Fort I intrusion osteotomy is usually the treatment of choice.<sup>23,24</sup> A surgical expansion of maxillary dental arches in a multisegment Le Fort I osteotomy is often followed by a certain amount of relapse.<sup>25–27</sup> It was reported that transverse stability after multisegment Le Fort I osteotomies could be improved when rigid internal fixation was applied and most relapse was noticed in the first year after surgery.<sup>27</sup>

In the case study at hand, presurgical orthodontics could be done, but with frequent rebonding of attachments and a one-piece Le Fort I intrusion osteotomy. Prosthodontic rehabilitation was completed 14 months after maxillofacial surgery with stable conditions.

# Adhesive Restorations of AI-Affected Teeth

For restorations of structurally compromised tooth substance-as is the case for AI-adhesive bonding techniques and tooth-toreconstruction bond properties must be at the center of attention. There are two possibilities for the prosthodontic rehabilitation of such teeth: (1) the use of gold or porcelain-fused metal crowns fixed with conventional cements (e.g., zinc-phosphate cements), or (2) the application of adhesive materials or fillings,<sup>28</sup> especially for the transitional treatment of the adolescent patient.<sup>29</sup> The advantage of conventional metal crowns lies in the high precision of workpieces and good long-term results; their drawback is the loss of tooth substance because of shoulder preparation.

Regarding bonding to enamel, one must consider acid-etched patterns. The typical chalk-white enamel surface after phosphoric acid etching is a sign that bonding to enamel can be established.<sup>30</sup> Because AI patients suffer from a reduced enamel layer (especially in hypoplastic AI), not only bonding to enamel but also to dentin is important. Bond strength to sclerotic dentin is well known to be less effective than to healthy dentin.<sup>31–33</sup> A study could show that dentin structure and mineralization in hypocalcified AI are changed with thicker peritubular dentin and dentin tubuli are partly or completely sclerotic.<sup>34</sup> Other types of AI with a reduced enamel layer must be expected to show the same or similar characteristics. Clinical signs for sclerotic dentin can be abrasiveness, glossiness, or discoloration. Short etching with phosphoric acid (total etch) or the application of self-etching primers is recommended.<sup>32</sup> Care must be taken when dentin hypersensibility is present.35

#### Cost and Long-Term Aspects

The cost of the orthodontic, maxillofacial, and prosthodontic treatment may be considerable, but is often covered by basic health insurance. An interesting analysis showed that 27% of the initial treatment costs were generally needed for the long-term maintenance and treatment of the



Figure 13. Intraoral views after prosthodontic rehabilitation with all-ceramic crowns: A, frontal view, B, occlusal view of the maxilla, C, occlusal view of the mandibula, D, right side view, and E, left side view.



*Figure 14. Panoramic radiography at completion of treatment.* 

consequences of biological and/or technical failures and complications over a period of 16 years for young adults with birth defects.<sup>36</sup> More long-term information on outcomes after fixed reconstructions are given by Pjetursson and colleagues,<sup>37</sup> showing that complications usually only appear after 10 years of function or later. There are only two reports on fixed reconstructions with AI that systematically list failures and complications over long observation times. One of them indicates that all AI patients were affected positively by the received treatment and biological and technical failures are no higher than for comparable patients with no birth defects and conventional fixed reconstructions.<sup>19</sup> The other study reports that of five patients with AI, only one remained complication-free after 18 years, with the main reason of the loss of 26 out of 92 crowns being purely esthetic.<sup>38</sup>

#### CONCLUSION

The multidisciplinary treatment of AI with AOB is a challenging task that can successfully restore functional and esthetic deficits caused by the AI. In the presented case, the main goal was to close the AOB, to reduce dentin hypersensibility, and to provide good facial esthetics by all-ceramic crowns. To ensure long-term success, a strict recall system and good oral hygiene are important.

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Figure 15. Final facial photograph at the age of 20.

#### DISCLOSURE STATEMENT

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