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Management of Amelogenesis Imperfecta: A 15-Year Case History of Two Siblings

E Dursun • E Savard • C Vargas • L Loison-Robert
H Cherifi • F Bdeoui • M-M Landru

Clinical Relevance

Restoring amelogenesis imperfecta–affected teeth with composite is a viable medium-term treatment in young adults and a conservative and esthetic approach that allows for repairs. Stainless steel crowns may be used to restore deciduous and adult molars. Adjusting crown molds using a thermoforming procedure and buildup of composite shells is an interesting technique for restoring affected premolars and canines.

SUMMARY

Objective: Amelogenesis imperfecta (AI) is a heterogenous genetic disorder that interferes with normal enamel formation in the absence of systemic disorders. The patients' main concerns are caries susceptibility, poor esthetics, and generalized sensitivity. There is a broad clinical spectrum, from discolorations to consequent enamel alterations. This case report describes the 15-year case study and the full-mouth rehabilitation of two siblings affected by a hypocalcified AI.

Clinical Considerations: In these two patients, conservative care with stainless steel crowns and direct composite restorations was undertaken to restore function and esthetics and to reduce sensitivities in primary and mixed dentitions. The difficulties in monitoring resulted in severe infectious complications (dental abscess with cutaneous fistula), important dental defects, and loss of spaces with subsequent malocclusion. In the young adult dentition, they were treated by extractions, root canal therapies, and new restorations: stainless steel crowns for permanent molars, direct composite restorations (with strip crowns) for

*Elisabeth Dursun, DDS, PhD, associate professor, Dental Materials Research Unit, Innovations and Interfaces (URB2i) and Pediatric Dentistry Department, Faculty of Dental Surgery, Paris Descartes University, Montrouge, France; Pediatric Dentistry Department, Albert Chenevier Hospital, Créteil, France

Elodie Savard, DDS, Faculty of Dental Surgery, Paris Descartes University, Montrouge, France; Restorative Dentistry and Pediatric Dentistry Departments, Albert Chenevier Hospital, Créteil, France

Carole Vargas, resident, Orthodontic Department, Rangueil Hospital, Toulouse, France

Ludwig Loison-Robert, DDS, assistant professor, Biology Department, Faculty of Dental Surgery, Paris Descartes University, Montrouge, France; Albert Chenevier Hospital, Prosthodontic Department, Créteil, France

Hafida Cherifi, DDS, assistant professor, Public Health Department, Faculty of Dental Surgery, Paris Descartes University, Montrouge, France; Oral Surgery Department, Albert Chenevier Hospital, Créteil, France

Fadi Bdeoui, DDS, hospital practitioner, Pediatric Dentistry Department, Albert Chenevier Hospital, Créteil, France

Marguerite-Marie Landru, DDS, PhD, associate professor, Pediatric Department Faculty of Dental Surgery, Paris Descartes University, Montrouge, France; Pediatric Dentistry Department, Albert Chenevier Hospital, Créteil, France

*Corresponding author: 1 rue Maurice Arnoux, Montrouge, 92120 France; e-mail: elisabethdursun@gmail.com

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Figure 1. *Intraoral views of the initial situation for the sister in primary dentition (3 years old) affected with a hypocalcified form of AI.*
Figure 2. *Intraoral views of the initial situation for the brother in primary dentition (6 years old) affected with a hypocalcified form of AI.*

incisors and maxillary canines (to improve the crown morphology as well as to mask the discolorations and the malpositions), and adjusted composite crown molds using a thermoforming procedure for premolars and the mandibular canines. The main difficulties were rapid tooth surface loss, bonding to atypical enamel, developing dentition, long-term follow-up.

Conclusion: Restoring function and esthetics in AI-affected patients is a challenge from primary to adult dentition. Early corrections are essential to avoid dental damage and for psychological benefits. This clinical report highlights the adhesive rehabilitation for anterior and premolar areas and the difficulty of patient follow-up.

INTRODUCTION

Amelogenesis imperfecta (AI) is an inherited and clinically heterogeneous disorder of tooth development that could affect all (or nearly all) primary and permanent teeth. Its prevalence ranges from 1:700 to 1:14,000, according to the studied population.[1] Three types have been described: hypocalcified AI (with soft and friable enamel), hypoplastic AI (with normally calcified enamel but deficient in quantity), and hypomaturation AI (characterized by mottled enamel, with opaque white to red-brown discoloration).[2] Furthermore, several other dental anomalies have been associated with AI, such as taurodontism, multiple impacted teeth, congenitally missing teeth, and malocclusions (open bite or class III).[3-5]

Rehabilitation is complex and requires a multidisciplinary approach. The treatment objectives are to reduce sensitivity and caries susceptibility, protect the tooth structure from wear, establish good oral hygiene habits, and restore esthetics and function to improve the patient's quality of life. The multiple treatment phases last several years, and thus the patient's compliance is essential. Different treatment options have been proposed for the management of AI-affected teeth: microabrasion, laminate veneers, composite resin restorations, composite or ceramic onlays, stainless steel crowns, and metal-ceramic/all-ceramic crowns.[6-10]

Nowadays, the treatment approach is rather based on dental tissue preservation. Recent studies have successfully used composite restorations,[7] and some authors even showed that these offered good long-term performance in permanent hypomineralized molars.[11] The aim of this article is to report the 15-year case study of two siblings affected by the hypocalcified form of hereditary AI.

CASES PRESENTATION

During Primary and Mixed Dentition

Two siblings were referred to the Albert Chenevier hospital of Créteil in 2000 for the treatment of discolored and sensitive teeth. Both were diagnosed with hypocalcified form of hereditary AI (Figures 1 and 2). Neither the parents nor the two eldest children seemed to be affected.

The 6-year-old brother was treated first with stainless steel crowns on the primary molars and composite restorations on the primary maxillary anteriors using strip crowns (technique described later in the article; Figure 3). When he was 7 years old and then 9 years old, he received stainless steel

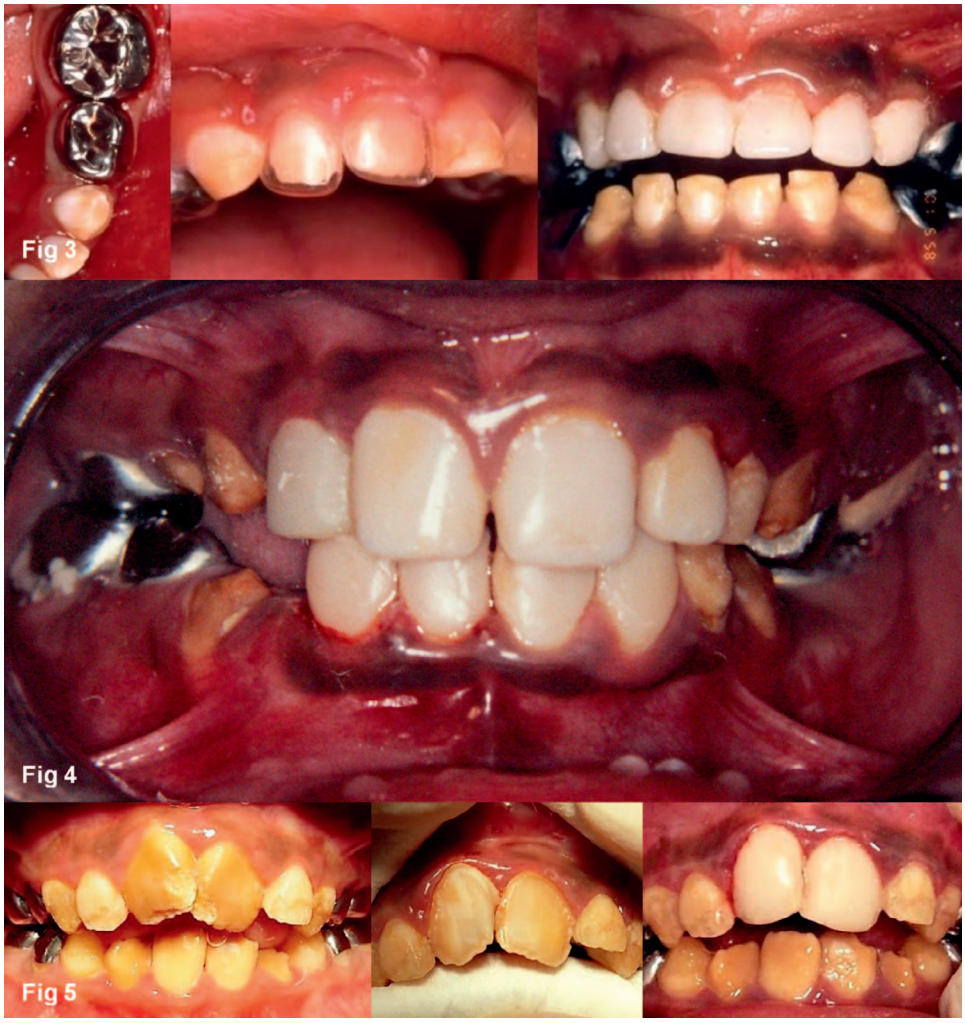

Figure 3. *Placement of stainless steel crowns on the boy's primary molars and composite restorations of the maxillary anterior primary teeth using strip crowns (6 years old).*
Figure 4. *Placement of stainless steel crowns on the boy's first permanent molars and labial composite restorations on the permanent incisors (between 7 and 9 years old).*
Figure 5. *Labial composite restorations of the sister's permanent maxillary central incisors (5 years old).*

crowns on the first permanent molars and direct composite restorations on the permanent centrals (Figure 4). At the age of 11, his maxillary premolars were covered with composite resin. As for the nearly 3-year-old sister, she showed no cooperation and thus treatment was not possible until she reached the age of 5, at which time the primary molars were restored with stainless steel crowns. When she was 9, a thin layer of composite resin was applied on the labial surfaces of the permanent maxillary central incisors (Figure 5). These patients did not show up for regular follow-up but rather for symptomatic treatments.

During Permanent Dentition at Adolescence

Case 1: The Sister—In 2013, the 14-year-old girl presented to the dental clinic, concerned about a chin fistula. She also complained of persistent tooth sensitivity to air, water, and brushing, as well as bad esthetics due to tooth discolorations.

Clinical Examination—Extraoral examination showed a slight facial asymmetry (nose deviation to the left) and a reduction in the dimension of the lower facial third. The cutaneous fistula was centered in the submental area. The 5-mm-wide lesion had a yellowish crust aspect, surrounded by an erythematous area (Figure 6). Intraoral examination revealed a red and swollen gingiva, heavy plaque deposits, as well as decayed, worn out, and discolored dentition (Figure 7). The old composite resins were no longer adapted to the gingival margins due to facial growth. An interincisal midline shift toward the left was noticed as well as a narrowed maxillary arch that caused an end-to-end molar relationship.

Radiographic Examination—The periapical radiographs revealed a large periapical radiolucency related to the mandibular right central incisor, which turned out to be the origin of the chin fistula. The bitewings showed several superficial radiolucencies related to the premolars and molars proximal

Figure 6. *Cutaneous fistula on the girl's chin (14 years old).*

areas, except for the second maxillary left molar, where the cavity was extending deep toward the pulp. The panoramic radiograph showed the impaction of the two maxillary permanent canines as well as the developing third molars. Cone beam computed tomography (CBCT) clarified the extent of the periapical radiolucency (which was approximately 10 mm wide) and the perforation of the external cortical bone next to the mandibular right central incisor. The impacted canines were positioned oblique and very close to the roots of the lateral incisors and first premolars (Figure 8).

Treatment Procedures—A treatment plan was elaborated to eliminate dental infections, reduce sensitivities, and improve esthetics and masticatory functions. The first steps were enhancement of oral hygiene and dental scaling. Endodontic treatment of the mandibular right central incisor and extraction of the severely decayed second maxillary left molar were carried out.

The preparation of the molars was as minimal as possible, especially on the occlusal surface, in order to compensate for the reduced vertical dimension. Stainless steel crowns were sealed using glass ionomer cement (Ketac Cem Radiopaque, 3M ESPE, Cergy Pontoise, France; Figure 9). The anterior teeth (incisors and retained deciduous canines) were restored in composite resin with strip crowns (transparent celluloid crown forms, Pella-ODUS). A slight circumferential preparation was performed to provide enough space for the strip crowns and the composite material, as well as to

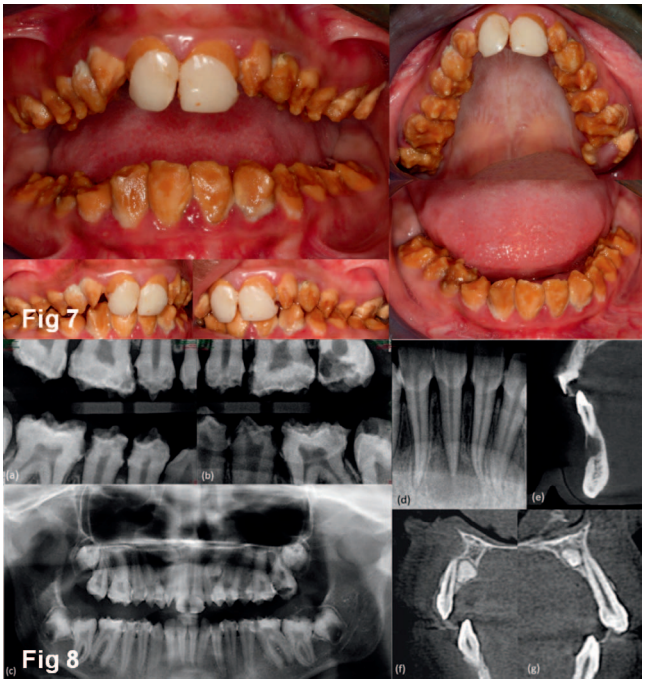

Figure 7. *Intraoral views of the sister's permanent dentition (14 years old).*
Figure 8. Radiographic images. (a, b) The bitewings reveal several superficial caries related to mandibular molars and premolars and deep cavity in the second maxillary left molar. (c) The panoramic radiograph shows impacted maxillary canines. (d) Large periapical radiolucent area next to the mandibular right central incisor. (e) Perforation of the external cortical bone (f). (g) CBCT examinations showing the impacted maxillary canines, close to the adjacent teeth.

remove the brownish-orange discolored surface layer, improving esthetic integration. The suitable size of strip crown was selected and trimmed with fine scissors to fit the tooth in length and cervical adaptation. The entire tooth surface was etched with a 37% phosphoric acid gel (Scotchbond Universal Etchant, 3M ESPE) for 30 seconds. The etchant was thoroughly rinsed, and the teeth were dried and subsequently treated with 5% sodium hypochlorite (NaOCl) for 1 minute to enhance the bond strength of the composite. Thin layers of primer and bonding agent (OptiBond, Kerr, Créteil, France) were then applied and light cured. The strip crown was filled with microhybrid composite resin (Herculite XRV, Kerr), carefully positioned on the tooth and the excess removed. After light curing for 80 seconds on each surface, the strip crown was peeled off with a probe or cut on the lingual side with a polishing disc if needed. Particular care was taken in the finishing and polishing sequence (using fine diamond burs, polishing discs, silicone cups), especially in the cervical region to avoid gingival inflammation. This procedure was repeated at a rate of two contralat-

Figure 9. *Minimal molar preparation and placement of stainless steel crown.*
Figure 10. *(a) Strip crowns adjusted on the prepared incisors. (b) Strip crowns filled with composite resin positioned on the prepared teeth. (c) Anterior teeth restored with composite crowns using the same technique.*

eral teeth per treatment session, starting from the central incisors (Figure 10). Therapeutic abstention was decided regarding the maxillary impacted canines. Thus, the two deciduous canines were restored using strip crowns and subsequently bonded to a wire splint fixed to the palatal surface of the adjacent teeth. The impacted permanent teeth were to be followed up regularly.

Given that no suitably sized strip crowns were available for the premolars and the mandibular canines, adjusted crown molds were produced using a thermoforming procedure. The maxillary and mandibular teeth on the same side were prepared and full arch impressions were taken. A thick layer of self-etch adhesive (Adper EasyBond, 3M ESPE) was applied on the reduced teeth to prevent sensitivity and protect the dental tissues until the subsequent appointment, scheduled no more than two days later. The plaster casts were mounted on a semiadjustable articulator (Quick Master) and waxed up. Partial alginate impressions of the wax-ups were made and poured in plaster for the subsequent thermoforming procedure. The casts were trimmed as close as possible to the tooth crowns. Transparent plastic sheets were heated and pressed onto the casts using a vacuum thermoforming machine (Original Essix 110V). After allowing the thermopressed sheets to cool down for a few minutes, they were removed and trimmed so as to obtain the crown molds. On the initial model, the built-up wax was removed and light-cured individual composite shells were created for each tooth (Figure 11). In the following appointment, the patient's teeth were polished to remove the adhesive layer, and the composite shells were tried in for proper fitting and contouring and then bonded with flowable composite (Tetric EvoFlow, Ivoclar Vivadent, Saint-Jorioz, France) of the same shade. Polishing was performed as described above, and the occlusion contacts were verified.

The patient as well as her family were very satisfied with the treatment outcome (Figure 12). The dental sensitivity was relieved, and masticatory function, esthetics, and social life were improved. The patient and her parents were informed that these treatment options were a good compromise as a transitional restorative alternative until completion of bone growth, but once the latter is achieved, readjustment or replacement of several restorations will be needed. The patient was monitored at a 3-month interval for 18 months. The restorations remained intact with a satisfactory surface quality and no recurrent caries (Figure 13). The permanent mandibular right second molar began to erupt. As for the root canal therapy of the mandibular right central incisor, the two-dimensional and three-

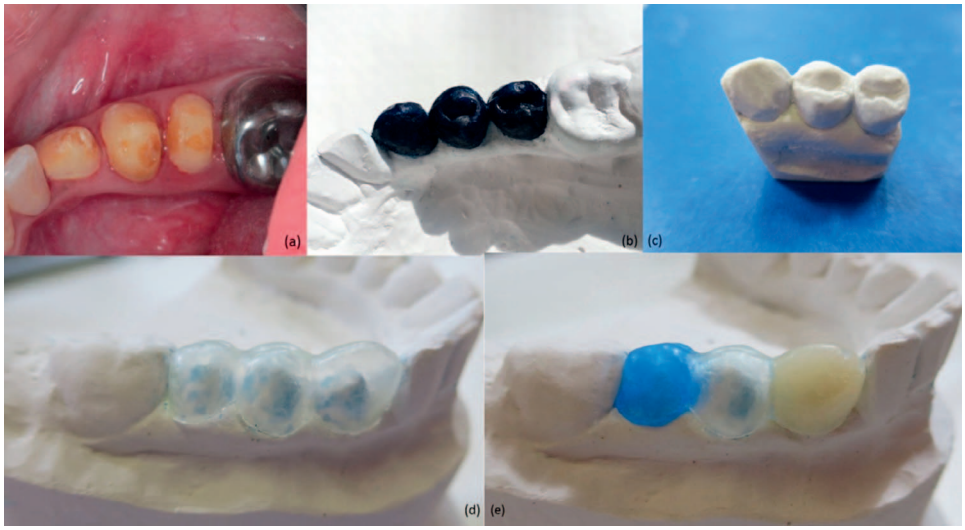

Figure 11. *(a) Mandibular canines and premolar preparations. (b) Wax-ups on plaster cast. (c) Reduction of the cast. (d) Crown template obtained using the thermoforming procedure. (e) Second individual composite shell elaboration thanks to wax isolation.*

dimensional radiographs showed a nearly complete regression of the radiolucency denoting bone regeneration. The chin fistula had completely disappeared (Figure 14).

Case 2: The Brother

While treating the young girl in 2013, her 18-year-old brother was encouraged to make an appointment for a checkup. He reported the persistence of mild tooth sensitivity since the last treatments in 2006.

Clinical Examination—Extraoral examination showed equal facial thirds, but an asymmetry was attributed to chin deviation to the left. Intraoral examination revealed heavy calculus deposits associated with moderate gingivitis. The second molars (except for the mandibular right one) were badly broken down. The stainless steel crown of the first mandibular left molar and all the composite restorations were no longer adapted to the gingival margins (Figure 15). The deciduous maxillary right canine showed high mobility. The absence of the contralateral canine was compensated for by the migration of adjacent teeth. Cross-bite on the left side, lateral infraocclusion on the right side, and edge-to-edge anterior bite were observable.

Radiographic Examination—The bitewings confirmed the maladaptation of the stainless steel crown on the first mandibular left molar. The periapical radiographs revealed a well-defined periapical radiolucency related to the maxillary left central incisor. The panoramic radiograph (Figure 16) showed the impaction of the two maxillary permanent canines and the mandibular third molars. According to CBCT sections, the impacted canines appeared close to the roots of adjacent teeth and generated internal and external cortical bone fenestrations. The third molars were in horizontal submerged position, in contact with the inferior alveolar nerve.

Treatment Procedures—Oral hygiene was enhanced and dental scaling performed. The maxillary left central incisor was endodontically treated. The broken-down second molars were extracted, as well as the impacted mandibular third molars, which led to perioperative pain and important postoperative complications for the mandibular left one (hemorrhage, trismus, bruise, and significant edema). The loose deciduous canine exfoliated by itself.

Regarding the mandibular left molars, the maladjusted crown was removed, the teeth were minimally prepared, and two new stainless steel crowns were simultaneously cemented.

The anterior teeth were restored with composite resin using strip crowns as described above. The designed wax-ups highlighted the impossibility of replacing the exfoliated deciduous canine because of insufficient space. Thus, it was decided to keep a narrow gap between the maxillary right lateral and first premolar. The premolars and the mandibular canines were restored following the same protocol described for the sister. Finding compromises for the buildup of composite restorations despite the malocclusion was challenging as the patient refused any orthodontic pretreatment.

At the end of the treatment, the patient was satisfied with the esthetic outcome and felt comfortable with his new occlusion. Follow-up was completed every 3 months for 12 months, during which oral hygiene motivation was always necessary. Except for a composite fragment that chipped off the buccal

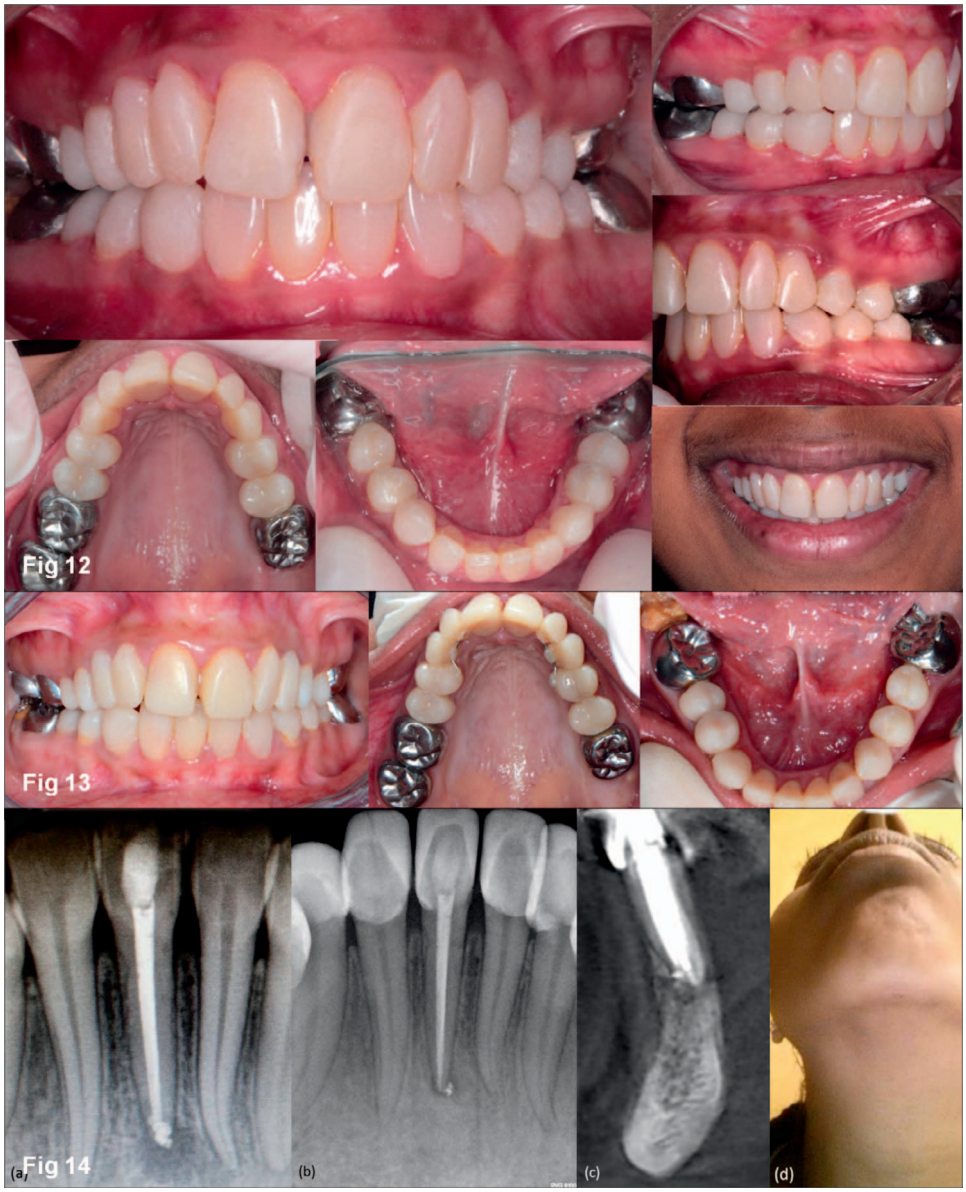

Figure 12. *Final intraoral views and smile photographs of the sister at the end of the treatment.*
Figure 13. *Intraoral views 18 months postoperative.*
Figure 14. *Radiographs showing the reduction of periapical radiolucency next to the mandibular right central incisor (a) 6 months after the endodontic treatment, (b, c) 18 months after the endodontic treatment, (d) skin healing of the fistula 6 months after the endodontic treatment.*

surface of the second left maxillary premolar at 7 months, the restorations remained satisfactory. After full eruption, the left maxillary third molar received a stainless steel crown (Figure 17).

DISCUSSION

Treatment Choice

Restoring esthetics and function in AI-affected patients is a challenging and multidisciplinary work and often requires compromises. Several therapeutic options were suggested: onlays, preformed stainless steel crowns, metallic or ceramic crowns for posterior teeth, and direct/indirect composite restorations, veneers, and ceramic crowns for anterior teeth. The treatment plan depends on numerous factors, including the type and severity of the enamel defects, the dentition stage, the hard and soft tissue development, and the cooperation and socioeconomic status of the patient.[12]

In primary and mixed dentitions, stainless steel crowns for primary molars and strip crowns or direct composites for anterior teeth remain efficient restorations and were chosen for the siblings' case. For permanent dentition, ceramic crowns and/or veneers are esthetically the more reliable solutions but are invasive, expensive, and require mature soft tissue. The treatment plan for these cases was based on the periodontal immaturity (which excluded fixed prosthodontics), the need of an acceptable esthetic

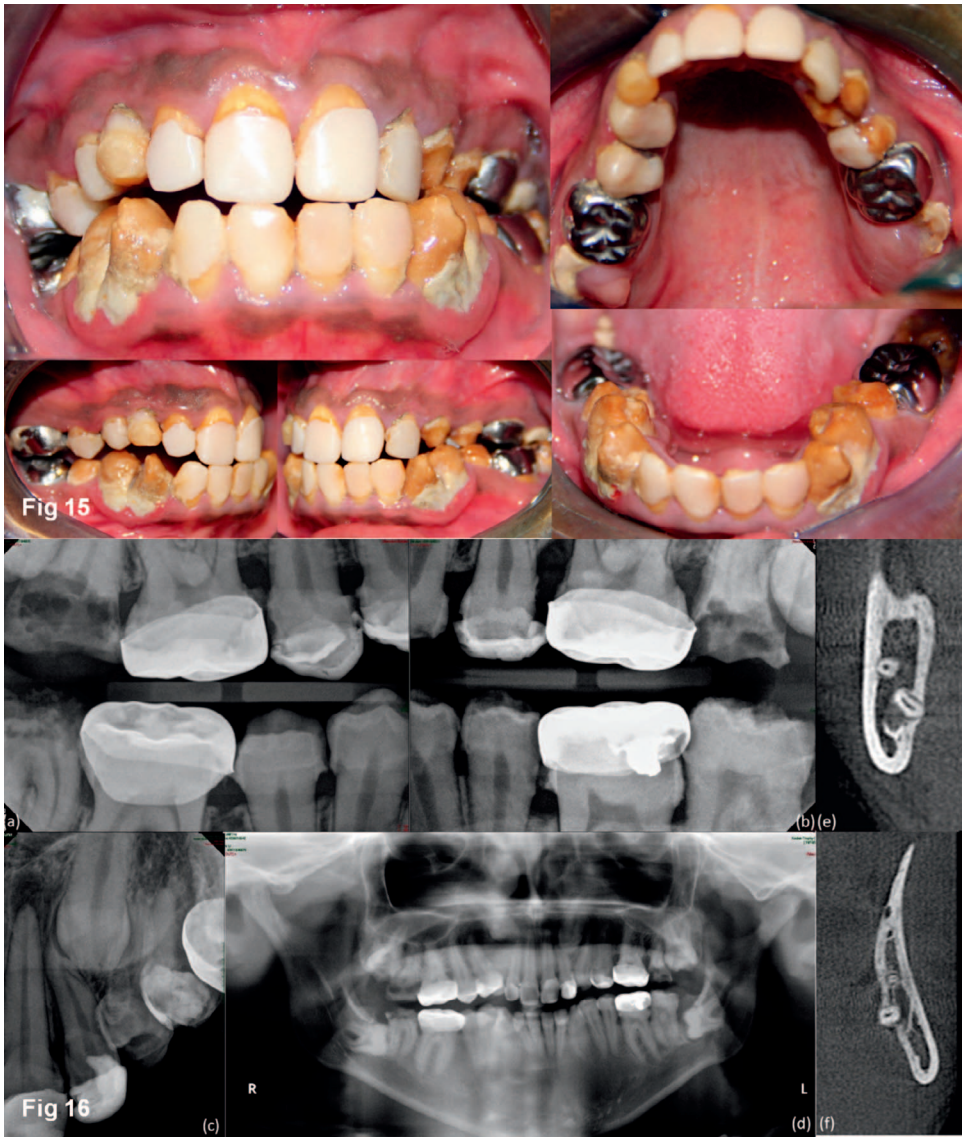

Figure 15. *Intraoral views of the brother's permanent dentition (18 years old).*
Figure 16. *Radiographic images. (a, b) The bitewings show the maladjustment of the stainless steel crown on the mandibular first left molar. (c) Periapical radiolucency next to the maxillary left central incisor. (d) Panoramic radiography showing impacted maxillary canines and mandibular third molars. (d, e) CBCT examinations of the impacted third molars showing their contact with the inferior alveolar nerve.*

appearance (requiring esthetic material), the relief of tooth sensitivity (indicating coverage of the entire crown surface), and the lack of financial resources of the patient's parents. These considerations led to the exclusion of ceramic crowns and veneer solutions. Composite restorations seemed to be a good compromise—esthetic and minimally invasive—as transitional restorations until bone and soft-tissue maturation. The crown molds for premolars and mandibular canines were also conservative, esthetic, functional, easily reparable, and affordable options. This transitional step allowed confirming the definitive form and shade for future prosthetic treatment.

Many studies reported a higher prevalence of impactions in AI patients.[10,13] In these siblings, the orthodontic traction of the impacted canines was not recommended as it might have led to root resorption of the adjacent teeth. Thus, these teeth will be subject to regular follow-ups. Their extraction will eventually be needed.

Bonding Composite Resin to AI Enamel

The conservative adhesive approach in young patients is more advantageous than the invasive prosthetic management.[14] In these cases, the bonding steps were difficult to perform because the siblings showed excessive salivation, and the placement of strip crowns or adjusted crown molds hindered the use of rubber dam or floss ligatures. Therefore, they needed to be performed by two practitioners and a dental assistant. Bonding to AI-affected enamel is more difficult compared with

Figure 17. *Intraoral views of the brother at 11 months postoperative.*

sound enamel, and adhesive restorations display higher failure rates.[15-17] The hypocalcified form of AI shows higher porosity due to widened interprismatic spaces and irregular and disorganized prism architecture.[18-20] It also contains a lower mineral content than sound enamel due to increased retention of amorphous organic material (enamel proteins) between the enamel rods.[21,22] The protein content would be about five times higher than in sound enamel,[23] which would result in significantly lower bond strength.[24]

To overcome this difficulty, it has been suggested to apply 5% NaOCl on the enamel surface prior to bonding, in order to remove excess enamel proteins and enhance bond strength.[24,25] However, other studies indicated that this deproteinization procedure had no significant effect.[17,26,27] As AI teeth frequently exhibit a thin and/or locally disrupted enamel layer, dentin bonding is generally involved as well. In the hypocalcified form, the underlying dentin is hypermineralized and displays a reduced number of tubules with narrow and partly obliterated lumen, surrounded by thickened peritubular dentin.[20,28] This morphological pattern corresponds to sclerotic dentin, on which bonding is reported to be less efficient.[29,30] According to some studies, dentin pretreatment with NaOCl would not negatively influence bonding agent effectiveness.[24,31] Thus, it was decided in these cases to apply 5% NaOCl on the tooth surface, after phosphoric acid etching.

Psychological Impact and Follow-up

The management of AI-affected patients requires the patients' and parents' motivation throughout the growth period, with successive and long treatment phases at each dentition stage. The treatment in permanent dentition lasted seven months. The regular follow-up was not easy to obtain, especially for the treatments in primary and mixed dentition, during which the patients missed several appointments and many treatment steps were left out. Finally, in these patients, as described in most cases,[32] the positive psychosocial impact was noticeable. After the rehabilitation, the two siblings had become more self-confident and smiled more.

CONCLUSION

This case report described the 15-year case study and in particular the recent treatment in permanent dentition of two teenage siblings affected by the hypocalcified form of AI. They underlined the challenges encountered from the primary to adult dentition. The rehabilitation was implemented with minimal tooth preparation: the permanent molars were restored with stainless steel crowns and all other teeth with composite resins. Eighteen months after the end of the last treatment in permanent dentition, all of the restorations were intact (without discoloration or infiltration around the margins) and the patients were very pleased with the results. The use of composite as a conservative treatment option is a viable alternative in the medium-term treatment of AI-affected dentition, to restore function and esthetics. Psychological motivation and patient cooperation play major roles in the advancement and outcome of the treatment procedures.

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Regulatory Statement

This study was conducted in accordance with all the provisions of the local human subjects oversight committee guidelines and policies of the Hôpital Albert Chenevier in Créteil, France.

Conflict of Interest

The authors have no proprietary, financial, or other personal interest of any nature or kind in any product, service, and/or company that is presented in this article.

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