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Clinical Technique/Case Report

Rehabilitation of an Adolescent with Autosomal Dominant Amelogenesis Imperfecta: Case Report

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Clinical Relevance

This case stresses the importance of preventive procedures in the restoration of dentition affected by amelogenesis imperfecta.

SUMMARY

Amelogenesis imperfecta is a hereditary condition that affects tooth enamel without systemic involvement. In the most severely affected patients, teeth can present alterations in enamel thickness, color and shape, all which compromise aesthetic appearance and mastigatory function. Several treatment options have been described to rehabilitate these patients, ranging from preventive intervention to a prosthodontic approach. Advances in the search for new techniques and bonding materials have provided less invasive treatment options. This study discusses the importance of preventive procedures and describes the clinical procedures of aesthetic and functional rehabilitation of a Brazilian adolescent with autosomal dominant amelogenesis imperfecta (ADAI) involving the use of direct and indirect resin composite restorations.

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INTRODUCTION

Amelogenesis imperfecta (AI) is a heterogeneous group of hereditary conditions that affects the quality and quantity of dental enamel without systemic involvement. It may affect all or some teeth in the deciduous and/or permanent dentition. X-linked, autosomal dominant and recessive modes of inheritance have been documented (Bäckman, 1997), and epidemiological studies have suggested AI to be an uncommon condition. In the United States, a prevalence of 0.06-0.07:1000 was reported (Wiktop, 1957). However, a ratio of 1.4:1000 was estimated in a northern Swedish County (Bäckman & Holm, 1986). These discrepancies may be due to diagnostic or demographic criteria or to mutant genes in the studied population. However, there is no available data to support the latter hypoth-

esis. With regard to the Brazilian population, no epidemiological data have been reported.

According to Wiktop's classification (1989), the defects of AI may be classified as hypocalcified, hypomaturation and hypoplastic types. The hypocalcified varieties are characterized by alterations in enamel mineralization. The enamel appears discolored and soft and can be easily removed. In the hypomaturation type, the enamel appears chalky and opaque. And, in the hypoplastic type, the defects are characterized by a deficiency in the quantity of enamel, which can be observed as pits, lines, grooves or thin enamel. Although this is the most widely used classification, AI presents a clinical heterogeneity that makes an accurate diagnosis difficult. Thus, a classification based upon the molecular defect, biochemical result, mode of inheritance and phenotype in the family involved seems to be the best alternative for future classification of AI cases and families with AI (Aldred & Crawford, 1995). Although recent research has identified several mutations of enamel proteins in X-linked AI and autosomal dominant AI (Forsman & others, 1994; Kärrman & others, 1996; Aldred & Crawford, 1997; Hart & others, 2000, Rajpar & others, 2001), the diagnosis rests mainly on the phenotype and mode of inheritance. An accurate diagnosis is necessary in order to determine preventive measures and adequate treatment.

Most of the problems caused by enamel fractures and wear and inappropriate invasive interventions can be avoided by early and correct diagnosis. Early management consists of medical advice and preventive dental care, such as dietary advice, oral hygiene instructions and topical fluoride application (Harley & Ibbetson, 1993; Mackie & Blinkhorn, 1991). In this context, the use of chemical agents that prevent biofilm formation could represent a valuable complement to mechanical plaque control.

Unfortunately, some patients only seek clinical evaluation when they present problems such as poor esthetics, teeth sensitivity, gingivitis and loss of occlusal vertical dimension. These are the most common problems related to AI patients (Rada & Hasiakos, 1990; Seow, 1993; Nel & others, 1997; Bouvier & others, 1999; Quinonez, Hoover & Wright, 2000). Often, these patients require a complex treatment and several options have been described for their rehabilitation. Treatment options include simple microabrasions (in cases of hypomaturation amelogenesis imperfecta) (Ashkenazi & Sarnat, 2000), NiCr crowns (Mackie & Blinkhorn, 1991; Harley & Ibbetson, 1993; Bouvier & others, 1999), gold or stainless steel crowns (Alexander, 1984; Harley & Ibbetson, 1993; Lumley & Rollings, 1993; Seow, 1993; Coley-Smith & Brown, 1996; Nel & others, 1997; Bouvier & others, 1999; Rosenblum, 1999; Quinonez & others, 2000; Yip & Smales, 2003), all ceramic crowns (Nel & others, 1997; Yip & Smales, 2003), ceramic metal crowns (Lumley & Rollings, 1993; Nel & others, 1997, Bouvier & others, 1999; Sari & Usumez, 2003; Yip & Smales, 2003), porcelain laminate veneers (Nel & others, 1997; Sari & Usumez, 2003; Yip & Smales, 2003; Gemalmaz & others, 2003), porcelain onlays (Nel & others, 1997; Gemalmaz & others, 2003), direct resin composite restorations (Harley & Ibbetson, 1993; Seow, 1993; Coley-Smith & Brown, 1996), indirect resin composite laminate veneers (Mackie & Blinkhorn, 1991; Yip & Smales, 2003) and indirect resin composite partial or full crowns (Quinonez & others, 2000). Clinical management may vary according to the degree of the defects and, in the most affected patients, treatment planning can be challenging.

In recent years, the use of adhesive materials has increased. In patients with defective teeth, resin composites are especially recommended for use directly or indirectly associated with glass ionomer cements, resin-modified glass ionomer cements and/or bonding agents (Croll, 2000). Besides the aesthetics they provide, resin composites preserve healthy dental structure and have good wear properties (Baratieri & others, 2001; Suzuki & others, 2002). Reported below is a case of aesthetic and functional rehabilitation of an adolescent with AI using bonding techniques with direct and indirect resin composite restorations.

CASE STUDY

A 16-year-old healthy Caucasian girl was referred to the Dental Clinic of the Hospital of the University of Brasilia, Brazil, for treatment of her pigmented, fragile teeth. According to the patient's mother, the deciduous teeth had also been affected but only to a minor degree. A complete dental and medical examination of the index case was performed and 12 family members were also examined. After prophylaxis, all teeth were dried,

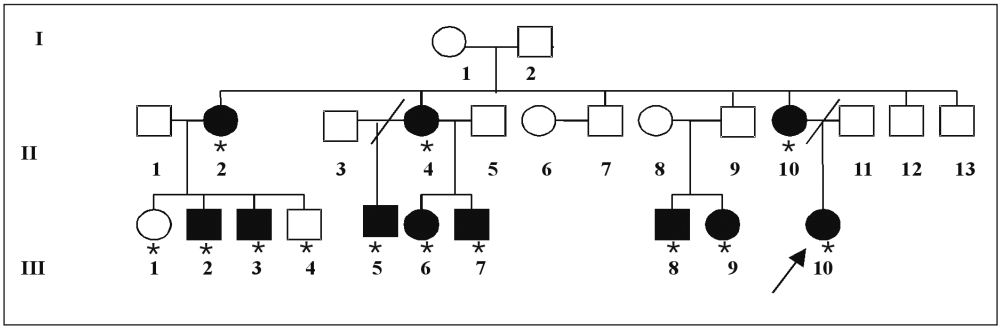

Figure 1. *The family pedigree shows that of 13 family members who were examined, 11 were affected. The asterisks indicate the clinically examined family members.*

and the defects were recorded according to the Developmental Defects of Enamel Simplified Index—DDE (FDI, 1992). Based on the genetic and clinical examination of proband and family members, a diagnosis of autosomal dominant AI (ADAI) was determined (Figure 1). The proband had both enamel hypoplasias and opacities in all teeth. Several family members had similar features, but others had either diffuse opacities or slight horizontal grooves (data not shown). Due to the clinical heterogeneity observed among members of the family, it was difficult to use the Witkop's classification (1989) to determine the type of AI, considering only the enamel defects.

Radiographic and Clinical Examination

An intra- and extra-oral radiographic exam was performed, and no significant difference in hard tissues contrast was verified. Oral hygiene was good and the gingival tissue was healthy. The clinical exam revealed the presence of hypoplasia in the form of grooves, lines, pigmented pits and areas with thin enamel. Also, there were diffuse opacities, teeth with shape and color alterations and areas with fractured enamel (Figures 2 and 3). The posterior teeth presented open cavities and fractured amalgam restorations with signs of microleakage (Figures 4 and 5).

The anterior teeth were fractured, and there was no available space to restore them. It appeared that, although the second molars did not show signs of wear, the premature wear and unsatisfactory restorations in the first molars had led to a premature loss of the occlusal vertical dimension and affected eruption of the second molars. Therefore, diagnostic casts were taken and mounted in central relation in a semi-adjustable articulator. A diagnostic wax-up was performed, and it was verified that the occlusal vertical dimension would have to be altered to allow for anterior teeth reconstruction. Also, the incisal and lateral guidances needed to be restored. It was estimated that about 1 mm high in the incisal pin would be necessary to restore the anterior and posterior teeth. In order to assess the supposed new vertical dimension, a phonetic test was performed, and the functional freeway space was evaluated (Tamaki, 1923). The new occlusal height seemed not to affect patient occlusion and function. Second molars were not included in the restorative procedures, but a passive eruption was expected to occur.

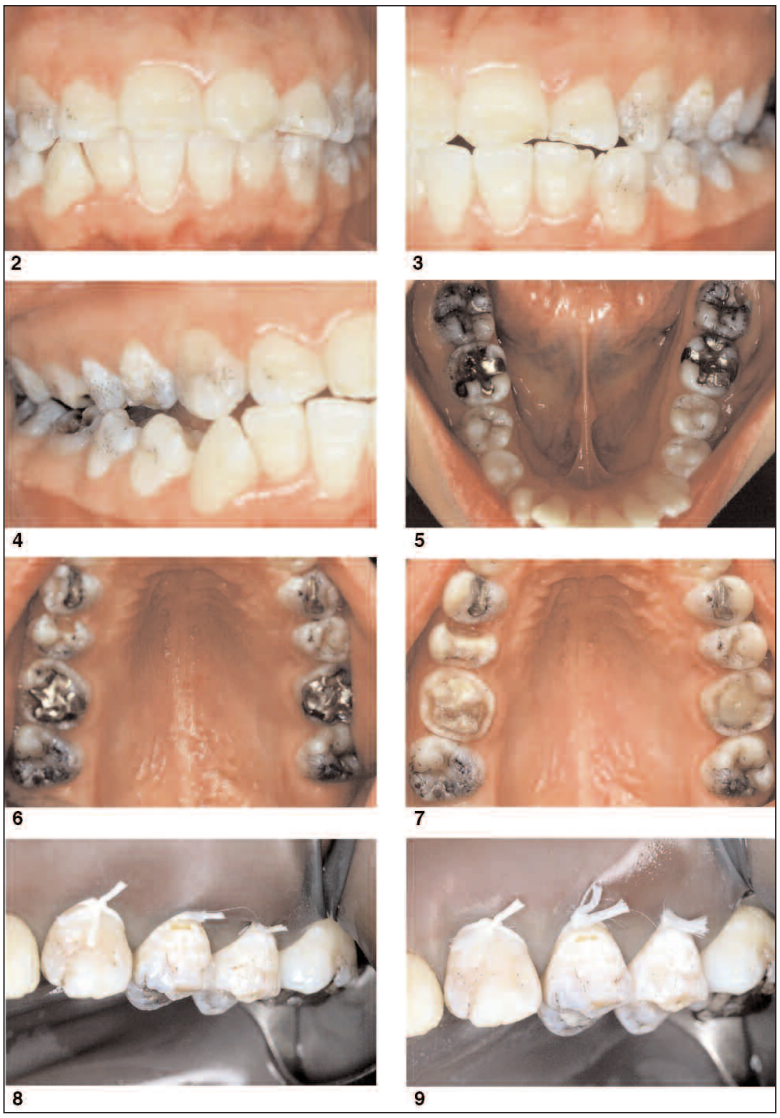

Figure 2. *Labial anterior view of the patient's pre-treatment smile, showing pigmented pits, grooves and fractured enamel areas with diffuse opacities.*
Figure 3. *Left labial view showing the patient's hypoplastic enamel and pigmented pits. Note that the lower incisors were not extrinsically pigmented and only exhibit an irregular enamel surface.*
Figure 4. *A right labial view showing the hypoplastic enamel and pigmented pits.*
Figure 5. *Occlusal view of the patient's lower posterior teeth presenting failed amalgam restorations and pigmented pits.*
Figure 6. *Occlusal view of the patient's lower posterior teeth showing the pigmented pits, failed amalgam restorations and fractured cuspal enamel.*
Figure 7. *Occlusal view of the patient's posterior teeth—the restorations were removed and the teeth were filled with glass ionomer and were prepared for impressions.*
Figure 8. *Left lateral view showing pigmented pits and wear in cuspal enamel.*
Figure 9. *Left lateral view showing the upper canine and premolars as they appear after aluminum oxide and sodium bicarbonate jet.*

After diagnosis, a three-phase treatment plan was proposed:

1) Preventive care
2) Restorative procedures
3) Case follow-up

Clinical Report

Since the patient presented good oral hygiene but had a cariogenic diet, preventive care involved oral hygiene instructions and nutritional advice. Topical fluoride application sessions were scheduled twice a year to prevent future lesions in the deep pits.

Regarding restorative procedures, direct and indirect restorations were indicated in accordance with the economic, esthetic and functional demands of the patient. The posterior tooth restorations were performed first in order to reestablish the occlusal vertical dimension. The indirect technique was chosen due to the extensive tooth destruction. The direct restorations were then executed to regain the aesthetics, overbite, overjet and incisal and lateral guidance according to the parameters established in the diagnostic wax-up.

The posterior teeth (15, 16, 26 and 36) (Figures 4 and 5) were restored with indirect resin composite restorations (Solidex, Shofu, Kyoto, Japan) to reestablish the occlusal vertical dimension and prevent future wear of the enamel. Previous unsatisfactory restorations were removed, and the teeth were completely filled with glass ionomer (Ketac-fil plus, 3M ESPE, St Paul, MN, USA). During a subsequent visit, tooth preparations were executed (Figure 6), impressions were taken with a polyether material (Impregum F, 3M ESPE) and provisional restorations (Protemp, ESPE, Seefeld, Germany) were placed with temporary cement (HydroC, Dentsply, Rio De Janeiro, Brazil). The casts were mounted in a semi-adjustable articulator. Once fabricated, onlays were tried-in and cemented with light-cured cement (Rely-X luting, 3M ESPE). The upper anterior teeth and upper and lower premolars, whose labial aspects were affected (Figures 2 and 3), were restored with direct procedures. Increments of resin composite were also added in the occlusal and incisal edges of these teeth to regain the incisal and lateral guidance, overbite and overjet. In spite of the presence of surface irregularities due to hypoplasia, the lower incisors were not esthetically affected (Figures 2 and 3); therefore, they were not restored, except for the lateral lower left incisor which was fractured at its distal incisal edge (Figure 3).

Similar veneer preparation was performed for the anterior teeth and premolars. The degree of tooth structure defect, intensity and deepness of color alteration and/or extrinsic pigmentation was considered as criteria of the stripping thickness. Abrasion was started on the labial surface by using a 40 mm aluminum oxide abrasive (Microetcher, Intraoral Sand Blaster, Danville Engineering Inc, San Ramon, CA, USA), followed by a sodium bicarbonate jet (Profi II, Dabi Atlante, SP, Brazil). This procedure removed a significant amount of pigmentation which had accumulated in the pits (Figure 8). The remaining pigmentation, which could interfere in the final esthetical result, was removed with small diamond burs (KG Sorensen, SP, Brazil).

For labial veneer preparation, a minimum chamfer was obtained with diamond burs (KG Sorensen). The proximal contact area was maintained and, incisally, no preparation was necessary, because the teeth heights were increased (Figure 9). Then, a 37% phosphoric acid etching was performed for 15 seconds and a bonding agent was applied (Single Bond, 3M ESPE). A thin layer of an opaque shade of composite (Charisma OA20, Heraeus Kulzer, Germany) was initially applied to mask the remaining spot (Figure 10), followed by a hybrid resin composite (Charisma color A20 and I, Heraeus Kulzer, Germany) and a microfilled resin composite (Filtek A110 color A2E, 3M ESPE, USA) (Figure 11). Resin composites from different manufacturers were used because of their availability at the Hospital of the University. The lower canines were restored with opaque and microfilled composites. The affected teeth, which were neither aesthetically involved nor decayed, were not restored, because the patient presented good oral hygiene.

Finishing and polishing were achieved with abrasive discs (Sof-lex/3M ESPE), 30-fluted carbide burs (KG Sorensen), abrasive flexicups, flexipoints, flexiwheels and polishing paste (Cosmedent Inc, IL, USA) (Figure 12).

The patient has been periodically recalled for clinical control and topical fluoride applications. After a 40-month follow-up period, the restorations were still in favorable clinical condition (Figures 13 through 18).

DISCUSSION

Treatment of patients with AI requires special care. These patients are sometimes psychologically affected due to the poor appearance of their teeth, and esthetical improvement can positively affect their self-confidence and behavior. In this case, this was clearly observed upon conclusion of the treatment.

Several reports have described the treatment of patients with AI. Clinical management can range from preventive interventions (Mackie & Blinkhorn, 1991) to complete rehabilitation (Lumley & Rollings, 1993; Nel & others, 1997; Bouvier & others, 1999; Sari & Usumez, 2003; Yip & Smales, 2003), depending on the severity of the case and when the diagnosis is made. Since this disease results from a hereditary condition, it is important to evaluate the dentition of the patient's relatives to aid in diagnosis. Thus, preventive procedures can be performed earlier, allowing defects to be correctly treated and future problems avoided.

In this case, the patient had already been submitted to several treatments with no preventive management. In general, little is known about both the etiology of AI

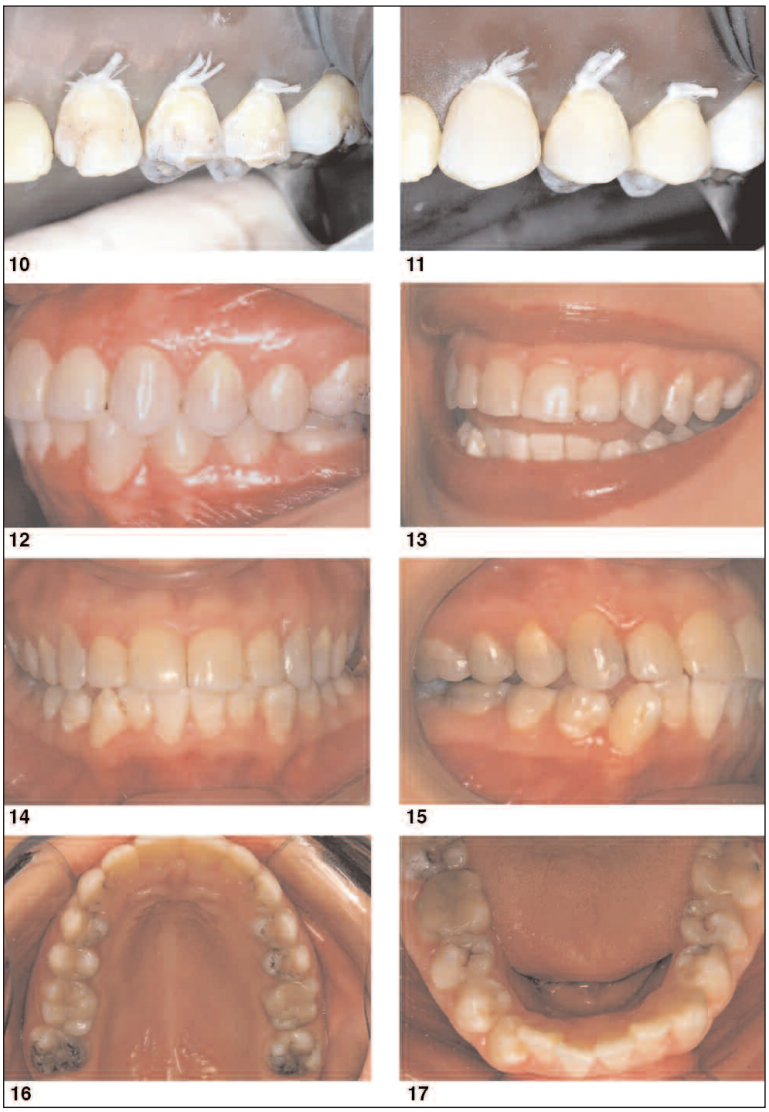

Figure 10. *Lateral view of finished preparations. Minimum wear with diamond burs provided space for the resin composite and removed a significant quantity of pigment.*
Figure 11. *Left lateral view after resin composite application and before polishing.*
Figure 12. *Left lateral view of the final esthetical result.*
Figure 13. *Labial view after of patient's post-treatment smile after 48 months.*
Figure 14. *Labial anterior view after 48 months.*
Figure 15. *Right lateral view: restorations are still satisfactory after 48 months.*
Figure 16. *Occlusal view of the upper posterior teeth. The second molars were not esthetically involved and were not restored (48 months).*
Figure 17. *Occlusal view of the lower posterior teeth. Direct and indirect restorations are still satisfactory after 48 months.*

and the appropriate dental materials used to restore tooth defects. An appropriate selection of restorative materials is important to preserve function and occlusal harmony and to restore the natural dentition morphology. Resin composites, glass ionomers, modified glass ionomer cements or a combination of these materials are recommended for restoring developmental dental enamel defects (Croll, 2000). Besides the esthetic properties of these materials, their adhesion to tooth structure is an advantage that may avoid the unnecessary removal of sound teeth.

Gold-based alloys have been commonly used to restore defective teeth (Lumley & Rollings, 1993; Nel & others, 1997; Bouvier & others, 1999). Although they display an unnatural appearance, they are wear-resistant (due to high tensile and compressive strength properties) and ensure minimal wear of the opposing enamel (Monasky & Taylor, 1971).

Despite of a greater esthetic potential, ceramics present a heightened abrasiveness to opposing tooth enamel, which, in some cases, is a limitation (Ratledge, Smith & Wilson, 1994; Hudson, Goldstein & Georgescu, 1995). This condition may be aggravated when the patient has fragile enamel due to developmental defects (AI, for example). In recent years, new low-fusing ceramic materials have been developed in an attempt to minimize wear damage (Imai, Suzuki & Fukushima, 2000). These low-fusing ceramic materials are significantly less abrasive to the opposing enamel than conventional porcelains, but gold-based alloys are less abrasive and more resistant to wear (al-Hiyasat & others, 1998).

The use of partial or full ceramic crowns has increased as a result of the demand for a natural-appearing restoration. However, poor prognosis has been reported for ceramic onlays, mainly because of their brittle characteristic, an inherent problem that affects their fracture susceptibility (Felden, Schmalz & Hiller, 2000). Thus, indirect resin composite materials have been used as good alternatives (Donovan & Kahn, 1990; Baratieri & others, 2001).

Recently, new materials called second-generation laboratory composites, poliglasses or ceromers have been developed. They have different filler components that improve wear resistance, physical properties (Rosenthal, Trinkner & Pescatore, 1997), and color stability (Donly & others, 1999); their use has expanded into posterior intracoronal, full crown and even fixed partial denture restorations (Freilich & others, 1998). Although the properties and wear characteristics of these composites are still significantly less desirable than those of gold alloys, these materials have superior properties: a good esthetical result, ease for repair, lower costs and a reduced abrasivity. Manhart and others (2000) observed, after a three-year evaluation of indirect resin composite restorations, that 93% of these restorations were assessed to be clinically excellent or acceptable (according to USPHS criteria).

Another concern in AI patients during treatment planning regards tooth sensitivity, a condition which is

commonly reported by these patients (Seow, 1993; Coley-Smith & Brown, 1996; Croll, 2000; Quinonez & others, 2000). Sometimes, full alloy crowns are indicated to eliminate sensitivity, prevent future wear of the dental structure and/or stabilize the vertical dimension (Alexander, 1984; Harley & Ibbetson, 1993; Lumley & Rollings, 1993; Seow, 1993; Coley-Smith & Brown, 1996; Nel & others, 1997; Bouvier & others, 1999; Rosenblum, 1999; Quinonez & others, 2000; Yip & Smales, 2003). In this case, the patient's posterior teeth presented extended areas with thin enamel and open cavities, but no sensitivity was reported.

Based upon all the factors discussed above, the authors opted to use indirect ceromer partial crowns in the posterior teeth. In the anterior teeth, in an attempt to avoid tooth preparation for full crowns or the classical preparation for labial veneers, the authors chose to reestablish the esthetic appearance and disclusal guidelines by using direct resin composite increments.

The management of severely affected patients can be challenging for the dentist. Innovations in adhesive and esthetic materials have opened up new horizons in treatment options. Advances in the field of adhesive materials have made restorative treatments less invasive, thus producing better esthetical results (Rada & Hasiakos, 1990; Croll, 2000). The patient's age and the quantity and quality of the affected enamel must be carefully evaluated in order to select the best treatment option.

Early diagnosis of amelogenesis imperfecta is the most favorable situation; however, when this is not possible, less invasive techniques should be chosen and the case should be followed up. A better understanding of AI is required in order to make a correct diagnosis. Therefore, early prevention measures and an appropriate restorative treatment may avoid iatrogenic treatments, which are sometimes observed in patients with AI. Additionally, more cases involving new materials and patient follow-up need to be reported in order to increase our knowledge of the clinical behavior of these materials in patients with AI. In this case, a four-year follow-up period showed that these materials were still in satisfactory esthetical and functional condition.

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