Oral Management of a Child with Mixed Dentition Affected by Amelogenesis Imperfecta

Ana Beatriz Silveira Moretti, DDS, MS Vivien Thiemy Sakai, DDS, MS Thais Marchini Oliveira, DDS, MS Sandra Harumi Fujiwara, DDS, MS Carlos Ferreira Santos, DDS, PhD Maria Aparecida Andrade Moreira Machado, DDS, PhD Salete Moura Bonifácio Silva, DDS, PhD

ABSTRACT

Amelogenesis Imperfecta (AI) is a hereditary developmental disorder affecting deposition, calcification or maturation of dental enamel in both the primary and permanent dentitions. Patients usually present tooth sensitivity as well as problems in chewing function and esthetics. In addition, dissatisfaction with the teeth appearance is often found, affecting the patient's social life. Oral rehabilitation of children with primary or mixed dentition is complex, since no definitive treatment can be done during periods of growth and until the end of eruption of the permanent dentition is accomplished. This clinical report describes the oral management of a 7-year-old girl with mixed dentition affected by AI. The patient showed accentuated teeth wear and root resorption, decreased occlusal vertical dimension, open bite and alteration in the sequence of eruption of permanent teeth. Most of the teeth were restored with resin modified glass ionomer cement. This approach markedly decreased the patient's dental hypersensitivity and improved functional chewing and esthetics. Positive psychological influence of the treatment on this patient was also observed. (J Dent Child 2007;74:157-60)

Keywords: Amelogenesis imperfecta, dental enamel defects, oral rehabilitation, glass ionomer

Melogenesis Imperfecta (AI) is a hereditary developmental disorder affecting dental enamel in both the primary and permanent dentitions.^{1,2} Four major types of AI were recognized based on the phenotype (hypoplastic, hypomaturation, hypocalcified and hypomaturation-hypoplastic with taurodontism) and subdivided into 14 subtypes based on phenotype and on mode of inheritance.^{3,4}

Al's major oral problems are extensive loss of tooth tissue, poor esthetics, and tooth sensitivity.^{1,5,6} Pulp and dentin are usually normal, and the teeth are usually caries resistant. AI has been associated with abnormalities in dental eruption, congenitally missing teeth, pulpal calcifications, root and crown resorption, hypercementosis, root malformations, taurodontism and open bite malocclusion.^{2,5-7} Despite the clinical features related to AI, patients are often affected by psychological problems.⁸ For favorable development of the stomatognathic system and also for psychological reasons afflicting children with AI, an early treatment strategy has been advised.⁹ This clinical report outlines the oral management of a girl with mixed dentition affected by AI.

CASE REPORT

A 7-year-old girl attended the Clinic of Pediatric Dentistry, Bauru School of Dentistry, University of São Paulo, Bauru, Brazil, with a request for dental care because of her esthetic problems and sensitive teeth. In the anamnesis, her mother reported that one of the patient's brothers and the father presented similar dental findings, and that the father had all his teeth extracted at an early age. Regarding her psychological condition, the patient suffered from a poor self-image due to the teeth appearance.

Clinically, the primary and permanent teeth were yellowish in color with rough enamel surface. The patient showed

Drs. Moretti, Sakai, Oliveira and Fujiwara are gradute students, Drs. Santos and Machado are associate professors, and Dr. Silva is assistant professor, all at Bauru School of Dentistry, University of São Paulo, Brazil. Correspond with Dr. Silva at samoboni@fob.usp.br

accentuated teeth wear, decreased occlusal vertical dimension, open bite and alteration in the eruption sequence of permanent teeth. Unerupted permanent first molars and premature mobility of primary teeth were also observed. Oral hygiene was not satisfactory, with evidence of gingivitis due to the accumulation of great amount of calculus and dental plaque (Figures 1-3).



Figure 1. Initial frontal view of mixed dentition affected by amelogenesis imperfecta in a 7-year-old girl. The patient showed accentuated teeth wear, open bite, great amount of calculus, dental plaque and gingivitis.

peared as a narrow vertical radiolucent channel interrupting the integrity of the crown (Figure 4).



Figure 4. Panoramic radiograph of a girl affected by amelogenesis imperfecta showing accentuated root resorption of primary teeth and an established communication between the pulp chamber and the tooth surface of the unerupted permanent maxillary first molars.

After thorough examination, the diagnosis of hypomaturation AI type was established, and the treatment plan developed taking into account the disorder's severity, the child's age and her psychological condition. At the beginning of the dental treatment the patient was uncooperative

> and did not permit a simple dental examination.

> sions, the primary maxillary left first molar exfoliated, and the primary maxillary right

lateral incisor was extracted.

Great discomfort was encoun-

Initially, scaling and root

planning of all teeth were performed. Dental plaque was disclosed, and the patient was taught how to improve her oral hygiene through better brushing techniques

Figures 2A and 2B. Maxillary (2A) and mandibular (2B) teeth affected by amelogenesis imperfecta. Preoperative view in a 7-year-old girl.



Figures 3A and 3B. Right (3A) and left (3B) lateral view of mixed dentition affected by amelogenesis imperfecta. Decreased occlusal vertical dimension and evidence of gingivitis due to the accumulation of great amount of calculus and dental plaque can be observed.

Radiographically, accentuated root resorption of primary teeth was detected, with no characteristics of pulp alterations and trauma. Radiographs of the unerupted permanent maxillary first molars showed an established communication between the pulp chamber and the tooth surface, which aptered during treatment, even after the administration of local anesthetics, due to the difficulty in achieving an adequate depth of analgesia.

The erupting permanent maxillary left central incisor was temporarily dressed with resin modified glass ionomer

and the additional use of a 0.12% chlorehexidine solution applied topically with a cotton swab after lunch and at bed time for 7 days. All the primary teeth were restored with resin modified glass ionomer cement (Vitremer, 3M ESPE, St. Paul, Minn, USA) after isolating the operating field with cotton rolls (Figures 5A and 5B). The teeth were not prepared with burs before its coverage with the restorative material. During the restorative ses-



Figures 5A and 5B. Primary maxillary (5A) and mandibular (5B) teeth restored with resin modified glass ionomer cement.



Figures 6A and 6B. Temporary dressing of the erupting permanent maxillary left central incisor with resin modified glass ionomer cement (6A). After the completion of its eruption, this tooth was restored with composite resin (6B).



Figure 7. Extracted permanent maxillary right central incisor due to coronal fracture.



Figure 8. Twenty two-month follow-up panoramic radiograph of a girl affected by amelogenesis imperfecta showing her oral status. The roots of the unerupted permanent mandibular first molars were completed and markedly curved distally.

cement (Figure 6A) in an attempt to provide support to the remaining healthy tooth structures, thus avoiding crown fracture. After it erupted, this tooth was restored with composite resin, providing improved esthetics and greater resistance (Figure 6B). The permanent maxillary right central incisor failed to erupt and, after ulectomy it began to erupt completely malformed. Unfortunately, due to the dental structure's fragility, this tooth fractured and had to be extracted (Figure 7).

After the entire restorative treatment, which took 20 months, the patient's dental hypersensitivity decreased markedly, and functional chewing was established. As a consequence of the oral rehabilitation, the patient presented a positive feedback regarding psychosocial

living, thus improving her quality of life. Twenty-two months after the beginning of the treatment, the roots of the permanent mandibular first molars were completed and markedly curved distally. These teeth, however, did not erupt spontaneously (Figure 8).

The patient has been followed for maintenance of oral health and for the evaluation of eruption of all permanent teeth. A definitive treatment with porcelain crowns shall be established when facial maturity has been reached.

DISCUSSION

Dental management of AI patients traditionally focuses on the restoration of the esthetics and function, which can be achieved with an approach of a dedicated team composed of the dentists, the patients and their parents. In adults or children with complete permanent dentition, a prosthodontic treatment can be performed with adhesive techniques, full porcelain crowns, porcelain-fused-to-metal crowns, overdentures, inlay/onlay restorations or crowns.² However, the oral rehabilitation of children with primary or mixed dentition is more complex, since no definitive treatment can be done during periods of growth and until the end of eruption of permanent dentition is accomplished.

Dental radiographs of AI teeth provide important information for the clinician regarding the degree of enamel mineralization, which enables the development of more appropriate treatment plans. All restorative treatment and materials for AI patients must be carefully considered in the context of the enamel mineral content. Restorations that require the invasion of existing enamel or bonding may be contraindicated for patients with AI who have thin or poorly mineralized enamel.⁶ Some authors^{5,10} reported that tooth enamel affected by AI can be treated by adhesively placed direct resin composite restorations. In the present case, composite resin and resin-modified glass ionomer cement were used, with the duration of the acid etching procedure similar to that indicated for normal teeth. The knowledge of the acid-etching patterns of the different clinical types of AI can help to prevent the high incidence of failure in the restorative treatments applied to these types of teeth.¹¹

Taking into account that a glass ionomer cement-based material tolerates better the presence of moisture, its choice as the restorative material of primary teeth appeared to be appropriated since rubber dam isolation could not be performed. Moreover, it allowed a decrease of teeth sensitivity, improvement of esthetics and fluoride release to oral environment.¹ Primary molars were restored in order to increase the occlusal vertical dimension and preserve the chewing function until the succeeding permanent teeth erupt. The rehabilitation of these teeth with stainless steel crowns was evaluated, but this approach could not be used due to the accentuated root resorption and to the possibility of accelerated teeth exfoliation.

Despite the early exfoliation of primary teeth with consequent quick eruption of premolars, the permanent mandibular first molars remained unerupted. The difficulty for the eruption of permanent first molars in AI patients was also observed by other authors.^{1,6,12} To establish an approach for the management of the multiple unerupted teeth, orthodontist help will be suggested in a near future.

Several reports have described an unusual malocclusion occurring in some AI patients that is characterized by failure of the maxillary and mandibular anterior teeth to meet in occlusion.^{27,9} This condition was also observed in our patient, but an orthodontic appliance could not be placed due to the great mobility and small cervico-occlusal length of the deciduous molars and premolars respective.

Two of the most important and valid reasons for replacing a missing incisor is to restore a natural and pleasing appearance and thus provide an opportunity for normal psychological development. In the present case, the construction of a fixed prosthesis to replace the extracted maxillary central incisor was not performed. This crown would have to be replaced as needed during the growth period to prevent a loss of space at the extraction site. Unfortunately, the mother could not afford the high costs of multiple replacements of the fixed prosthesis.

There are many manifestations of AI, and there is no standard formula for successful treatment. Sometimes an interdisciplinary approach may be needed.^{2,9} In our patient, the restoration of almost all teeth with resin modified glass ionomer cement satisfied the functional and esthetic expectation of the patient and dental team even though a definitive rehabilitation could not be performed. Besides, the satisfactory clinical result was only achieved due to the patient's awareness concerning the importance of keeping

oral health through adequate oral hygiene.

In conclusion, dentists should be aware of the presentation of amelogenisis imperfecta to assist in early diagnosis and aim to provide the patient with the proper oral rehabilitation treatment. It is worth mentioning that dental treatment can be necessary for medical reasons and has far-reaching implications for overall health of AI sufferers.⁸ Therefore, the social implications for AI patients should be considered and an early intervention to relieve their suffering is advised.

REFERENCES

- Lykogeorgos T, Duncan K, Crawford PJ, Aldred MJ. Unusual manifestations in X-linked amelogenesis imperfecta. Int J Paediatr Dent. 2003;13(5):356-61.
- Toksavul S, Ulusoy M, Türkün M, Kümbüloglu. Amelogenesis imperfecta: the multidisciplinary approach. A case report. Quintessence Int. 2004;35(1):11-4.
- 3. Witkop CJ Jr. Amelogenesis imperfecta, dentinogenesis imperfecta and dentin dysplasia revisited: problems in classification. J Oral Pathol. 1988;17(9-10):547-53.
- 4. Aren G, Ozdemir D, Firatli S, Uygur C, Sepet E, Firatli E. Evaluation of oral and systemic manifestations in an amelogenesis imperfecta population. J Dent. 2003;31(8):585-91.
- 5. Türkün LS. Conservative restoration with resin composites of a case of amelogenesis imperfecta. Int Dent J. 2005;55(1):38-41.
- Collins MA, Mauriello SM, Tyndall DA, Wright JT. Dental anomalies associated with amelogenesis imperfecta: a radiographic assessment. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 1999;88(3):358-64.
- Ravassipour DB, Powell CM, Phillips CL et al. Variation in dental and skeletal open bite malocclusion in humans with amelogenesis imperfecta. Arch Oral Biol. 2005;50(7):611-23.
- 8. Coffield KD, Phillips C, Brady M, Roberts MW, Strauss RP, Wright JT. The psychosocial impact of developmental dental defects in people with hereditary amelogenesis imperfecta. J Am Dent Assoc. 2005;136(5):620-30.
- 9. Ozturk N, Sari Z, Ozturk B. An interdisciplinary approach for restoring function and esthetics in a patient with amelogenesis imperfecta and malocclusion: a clinical report. J Prost Dent. 2004;92(2):112-5.
- 10. Vitkov L, Hanning M, Krautgartner WD. Restorative therapy of primary teeth severely affected by amelogenesis imperfecta. Quintessence Int. 2006;37(3):219-24.
- Sánchez-Quevedo MC, Ceballos G, Rodríguez IA, García JM, Alaminos M. Acid-etching effects in hypomineralized amelogenesis imperfecta. A microscopic and microanalytical study. Med Oral Patol Oral Cir Bucal. 2006;11(1):E40-3.
- 12. Noffke CEE, Chabikuli NJ, Nzima N. Impaired tooth eruption: a review. SADJ. 2005;60(10):422,424-5.