10.5005/jp-journals-10024-1321

CASE REPORT

Multidisciplinary Management of a Child with Severe Open Bite and Amelogenesis Imperfecta

Catherine Millet, Jean-Pierre Duprez

ABSTRACT

Aim: To present a case of multidisciplinary management and fixed rehabilitation of a young girl with amelogenesis imperfecta (AI), a severe open bite and occlusal instability.

Background: AI is a genetic disorder characterized by enamel malformations, disturbances in tooth eruption and significant attrition. Early diagnosis is essential, since rapid breakdown of tooth structure may occur, giving rise to acute symptoms and complicated treatment. As AI is frequently accompanied by unesthetic appearance, open bite deformity and malocclusion, a multidisciplinary approach is often required.

Case report: This clinical report describes the condition and presents the case of a 10-year-old girl with hypocalcified form of AI. Orthodontic treatment and orthognathic surgery were performed as part of the prosthetic treatment plan to achieve acceptable and durable results. They consisted of correcting class II, posterior crossbite and anterior open bite with a fixed orthodontic appliance, Lefort I osteotomy, bilateral mandibular ramus osteotomy and genioplasty. Prosthodontics treatment consisted of metal-ceramic crowns with low-fusing ceramic for good long-term results. No deterioration in the rehabilitation was found after 5 years of follow-up.

Conclusion: Complete restoration of severe AI is a long and complex process generally extending over several years.

Clinical significance: This article shows the important role of interdisciplinary approach to treating a patient with AI over a period of 8 years.

Keywords: Amelogenesis imperfecta, Hereditary disorder, Oral rehabilitation, Orthognathic surgery, Case report.

How to cite this article: Millet C, Duprez JP. Multidisciplinary Management of a Child with Severe Open Bite and Amelogenesis Imperfecta. J Contemp Dent Pract 2013;14(2): 320-326.

Source of support: Nil

Conflict of interest: None declared

INTRODUCTION

Amelogenesis imperfecta (AI) is a hereditary dental disorder that affects the amount, structure and/or composition of enamel. AI affects both primary and permanent dentitions, and exists independently of any other systemic anomaly.[1,2] AI prevalence is estimated to occur between 1:700 and 1:14,000; the inheritance pattern may be autosomal dominant, autosomal recessive or X-linked.[3,4] Six genes are known to cause AI: AMELX (amelogenin), ENAM (enamelin), MMP-20 (enamelysin), KLK4 (kallikrein-related peptidase 4), WDR72 (WD repeat containing domain 72) and FAM83H (family with sequence similarity 83, member H).[5,6] The classification proposed by Witkop in 1988 contains four types: Hypoplastic, hypomaturation, hypocalcified and hypomaturation—hypoplastic with taurodontism.[1] In the hypoplastic type, enamel is relatively well mineralized but thin with a yellow-brown coloration. In the hypocalcified type, enamel is formed in relatively normal amounts but soft and easily removed from the dentin. This most severe type is characterized by extensive fracturing and radiopacity similar to that of dentin in radiographs. In the hypomaturation type, enamel is soft and opaque with mottled areas. This type is considered to be benign manifestation of the hypocalcified type. Each type may be divided into several subtypes depending on the mode of inheritance as well as clinical, radiographic, histological and genetic features.[7]

Numerous clinical features can be associated to AI, including gingival hyperplasia,[8] eruption disturbances, significant attrition, dental sensitivity, pulp calcifications, root and crown pathological resorption.[9] Craniofacial features may also be present, such as constricted maxillary arch, reversed mandibular curve of Spee, and skeletal anterior open bite (AOB).[10,11] Major problems in AI patients are compromised esthetics and psychological impact.[12] In addition, these patients may face chewing function problems due to dental sensitivity and loss of occlusal vertical dimension (OVD) caused by attrition.[2,13]

The general aim of the treatment for patients suffering from AI is to improve dental esthetics and chewing function. Numerous treatments have been described in adults and children.[14-16] Treatment strategy includes consideration of patient's age, severity of the attrition, function, esthetics, orthodontic need, the periodontal condition and the socioeconomic status. Most clinical reports in the literature concern the restoration of dentitions affected by hypoplastic-type AI. In the most severe cases, an interdisciplinary team of specialists is usually required, including a pediatric dentist, an orthodontist, an oral and maxillofacial surgeon and a prosthodontist.[17]

The purpose of this clinical report is to describe the multidisciplinary management and fixed rehabilitation with metal-ceramic restorations of a young girl with hypocalcified AI, a severe open bite and occlusal instability.

CASE REPORT

History, Diagnosis and Treatment Planning

A 10-year-old girl was referred to the Pediatric Dentistry Department of the Hospices Civils of Lyon, France, by her orthodontist because of considerable sensitivity and severe discoloration of all erupted teeth. In the anamnesis, the patient reported difficulty with mastication and poor self-image due to tooth appearance. Her medical history was unremarkable. There was no evidence of systemic disease, nutritional deficiency, or drug treatments that may have affected dentition structure during development. The patient's hair, skin and nails were normal. The family history revealed that her father was affected by AI and had received overdentures when he was 28 years old. Patient has a 3 years young sister. Both sister and mother have normal teeth. Extraorally, patient's face was long and symmetric. No sign or symptom of temporomandibular joint dysfunction, including pain, opening limitation or clicking was observed. Predominant mouth breathing was noticed. Intraoral examination showed a mixed dentition; first molars and permanent incisors had erupted except the maxillary left lateral incisor. Oral hygiene was not satisfactory, with severe plaque accumulation and gingivitis. All erupted teeth, in primary and permanent dentitions had a yellowish discoloration, with many irregularities and porosities. Enamel was soft and has fractured from the crowns except in the cervical areas. Cusp structure was altered in the occlusal portion of molars due to altered posteruptive breakdown. Composite resin restorations had been previously placed on all permanent central incisors which presented an asymmetric gingival contour. Occlusal examination showed the presence of an AOB, a bilateral class II Angle molar relationship, and a left posterior crossbite involving the molars. Radiographic examination revealed no contrast between enamel and dentin. Enamel appeared thin, with an irregular surface. The clinical phenotype and family history suggested an autosomal recessive hypocalcified AI (Figs 1A to E).

Due to the complex needs of the patient, a multidisciplinary approach was necessary. Treatment strategy was defined in three phases. The initial phase consisted of oral hygiene instruction, improvement periodontal health and protection of teeth against sensitivity and wear. The purpose of the second phase was to correct malocclusion and improve masticatory function with orthodontic therapy and orthognathic surgery after puberty. The last phase consisted in prosthodontic procedure including complete crowns on all teeth to stabilize the results and improve patient's appearance. The patient and her parents were informed about the treatment plan and gave their informed consent.

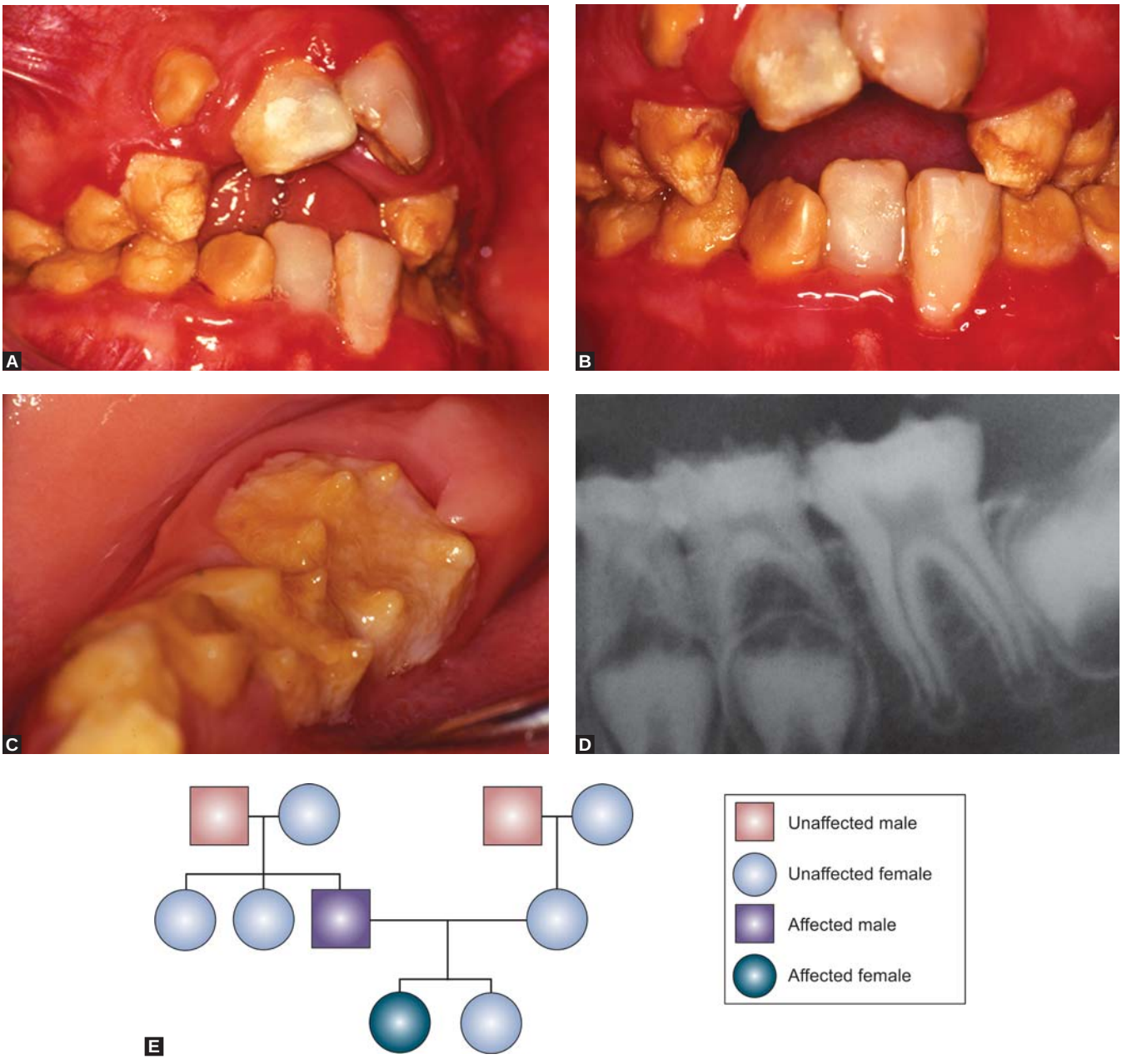
Treatment Procedures

Initial Phase

The first two appointments were directed toward obtaining full cooperation of the child for the predictably long and stressful sessions to come and toward motivating the child to adopt a positive approach to oral hygiene. During first four sessions, primary canines were extracted and preformed Ni-Cr crowns (Ion, 3M ESPE, St Paul, MN) were placed on first permanent molars and all primary molars to protect dentin-pulp complex, stabilize occlusion and decrease risk of loss of OVD in the future (Fig. 2). Upper and lower incisors were fitted with polycarbonate resin restorations (Ion, 3M ESPE) to improve appearance and decrease sensitivity. Patient's cooperation was good.

Second Phase: Orthodontic and Maxillofacial Procedure

Initial orthodontic treatment protocol in mixed dentition consisted in slow maxillary expansion using a Hyrax appliance (Fig. 3). The appliance was activated until an 8 mm expansion was obtained. The patient was monitored at 1 month interval to check for esthetic or functional problems and was encouraged to use the oral hygiene procedure established for her at home. Thereafter, the appliance was passively kept in place as a retainer. During this period, the child was monitored for permanent tooth eruption. First premolars were extracted. Second premolars and canines were fitted with carboxylate resin crowns, and second molars were fitted with Ni-Cr crowns. Pulp vitality was maintained for all teeth except for the maxillary left lateral incisor which required root canal treatment (RCT).



Figs 1A to E: Clinical photographs, radiograph and pedigree of the patient affected with AI

After teeth had been fitted, fixed orthodontic appliances were installed on mandibular and maxillary teeth to level and align dentition within each arch. After having reached completion of bone growth and 2 years of preoperative orthodontics, orthognathic surgery was performed to restore maxillary-mandibular dental relationship. So, at the age of 16, the patient underwent a midline split segmental Lefort I osteotomy in the upper jaw with impaction of 5 to 6 mm in the incisive region associated with an 8 mm posterior impaction. Additionally, a bilateral sagittal split osteotomy with genioplasty was realized to advance the mandible. The operation proceeded smoothly and without complication. After the consolidation period, the patient resumed orthodontic treatment, comprising closure of little diastemas and rotations. Postchirurgical orthodontic treatment lasted 12 months. In addition, maxillary and mandibular central incisors required RCT because orthognathic surgery caused their necrosis (Fig. 4).

Prosthodontic Procedure

When the child was 17.5, preformed crowns were removed and tooth preparations were completed for individual metal-ceramic crowns. New interim prostheses with mutually protected occlusal scheme were fabricated using autopolymerized acrylic resin (Unifast Trad, GC America, Alsip, IL) by direct technique. The patient wore the interim prostheses for 3 months, which allowed her to validate esthetics, function and comfort. Definitive impressions were

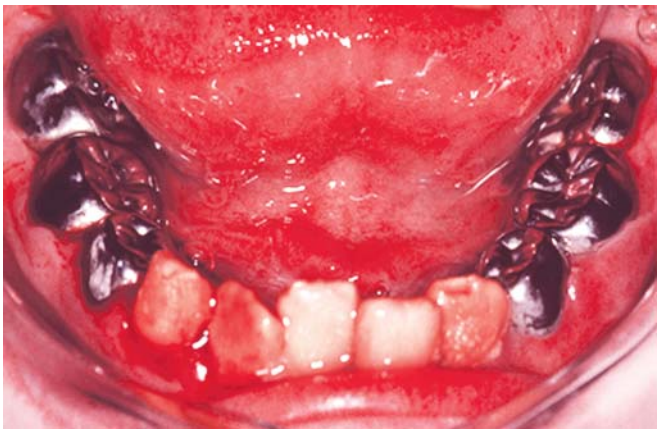


Fig. 2: Mandibular occlusal view showing preformed Ni-Cr crowns on primary and permanent molars

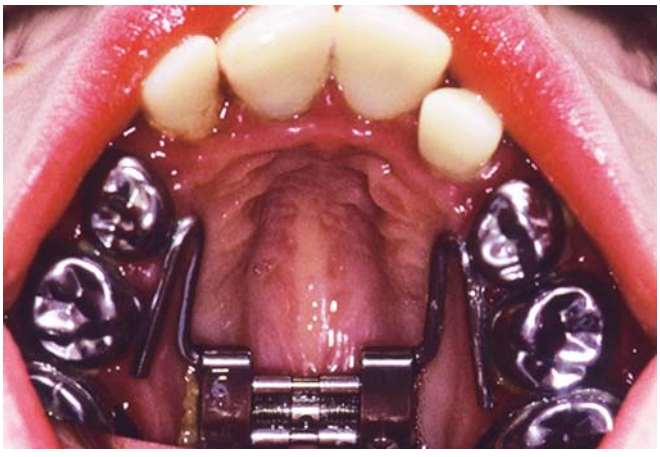


Fig. 3: Maxillary intraoral view during expansion

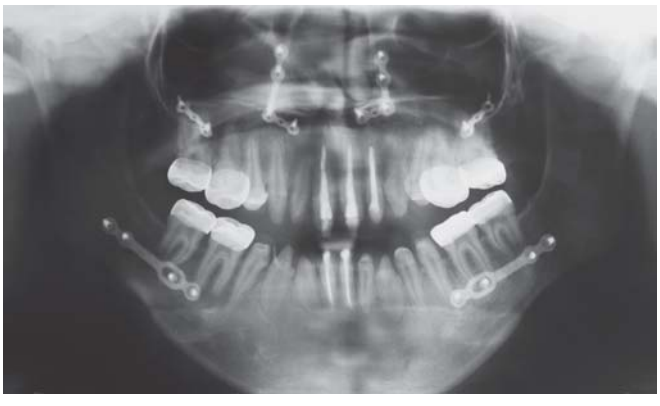


Fig. 4: Postoperative panoramic radiograph after maxillofacial surgery

made with individual impression trays using vinylpolysiloxane (VPS) (Express, 3M ESPE), and occlusal registrations were obtained with wax wedges (Moyco Industries Inc, Philadelphia, PA). Working casts were mounted on a semiadjustable articulator SAM 2 (SAM Präzisionstechnik, GmbH, Munich, Germany). Precious alloy (Degunorm, Degussa AG, Hanau, Germany) was used for producing all frameworks that were tried in the patient's mouth to check marginal fit and centric relation position. A low-fusing ceramic (Ducera, Degussa AG) was then applied to produce metal ceramic crowns on incisors, canines, premolars and mandibular molars. Both first and second maxillary molars were fitted with metal-ceramic crowns with metal occlusal surfaces. Occlusion was constructed as mutually protected scheme. A trial evaluation of the ceramic before final glazing enabled minor occlusal corrections. All restorations were cemented using glass ionomer cement (Fuji I, GC Dental Corp, Tokyo, Japan) (Fig. 5). The patient and her parents were very satisfied with the results (Fig. 6). There was a substantial occlusal improvement due to corrections of AOB and posterior crossbite. Class I canine and molar relationships were obtained on the right side, but the left side remained with a half-cusp class II relationship. Recall evaluations at 3-month intervals occurred for a period of 1 year. Gingival margins were stable with no inflammation. Evaluation 5 years after treatment showed good stability of both orthodontic and prosthodontic results (Figs 7A and B).

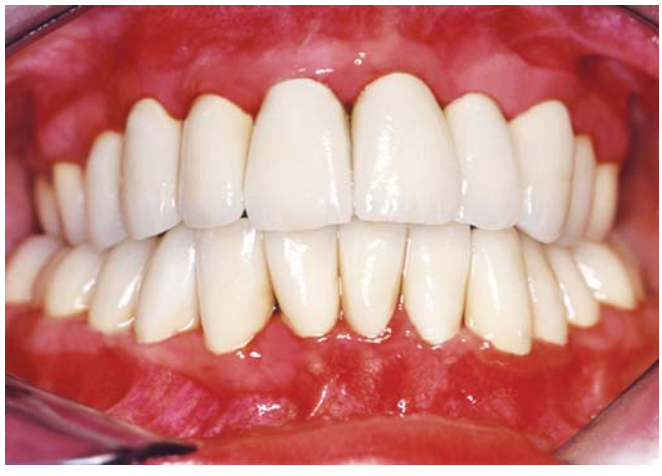


Fig. 5: Facial view of definitive restorations

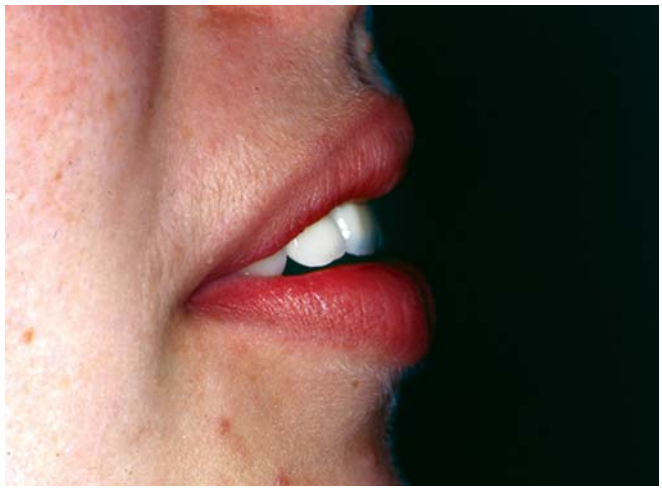
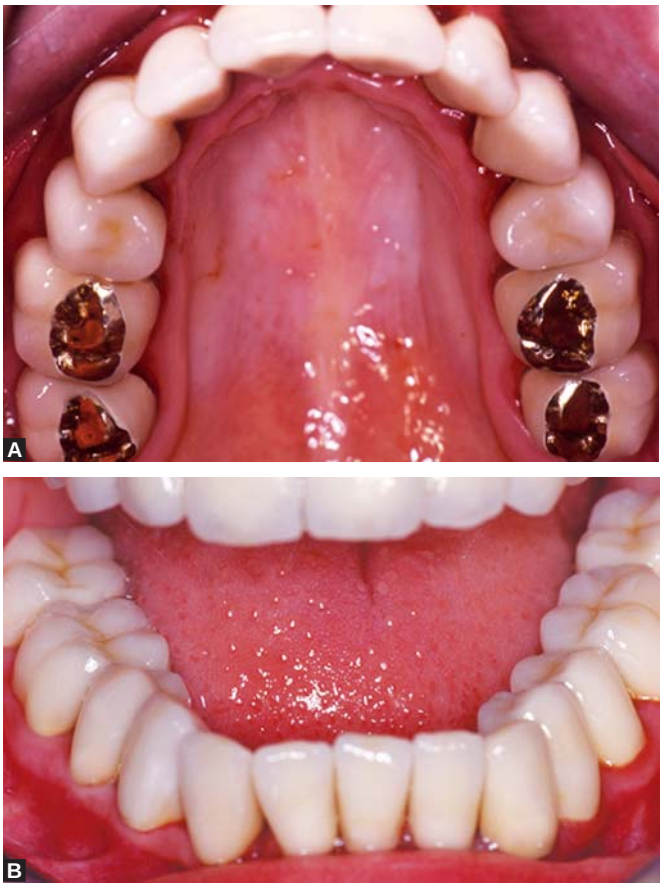


Fig. 6: Post-treatment extraoral view



Figs 7A and B: Intraoral views 5 years after insertion: (A) Maxillary occlusal view, (B) mandibular occlusal view

DISCUSSION

There is no standard strategy for successful management of AI. Early recognition followed by appropriate care is essential to prevent progressive damage of dentition and the psychological impact of the condition. It is a very long process that starts in the period of primary dentition. Craniofacial alterations occur with increased frequency in people with AI.[18,19] In most cases, posterior transverse discrepancies are due to insufficient maxillary arch width; therefore, treatment usually involves use of a maxillary, expansion appliance, which can take many forms. Ravassipour suggested that the etiology of open bite malocclusions in the different AI types is multifactorial, including abnormal eruption of posterior teeth because of gene mutation affecting both the dental and craniofacial developmental pathways or environmental factors.[11] Many orthopedic-orthodontic methods have been used to correct open bites.[20] In most patients, treatment requires a multisegment Le Fort I osteotomy with tilting of the occlusal plane.[10] Possible complications of multisegment Le Fort I intrusion osteotomy include periodontal defects and damage to roots adjacent to osteotomy cuts. Most relapse is noticed in the first year after surgery, and therefore, final occlusal rehabilitation should be postponed.[21] In this report, endodontic treatment of the four central incisors was necessary because their roots have been damaged by maxillofacial surgery. Prosthodontic rehabilitation was performed 18 months after surgery. Despite a great improvement in dental esthetics and occlusion, a compromised occlusal result was achieved. There is a normal overbite and overjet, but there is still a half-cusp class II posterior segment relationship on the left side. No occlusal changes were noted after 5 years.

In the present case, the clinical diagnosis was hypocalcified type AI. The low degree of enamel mineralization is likely to be one of the reasons for posteruptive breakdown.[22,23] That is the reason why, in mixed dentition, molars were restored with stainless steel crowns to preserve the OVD. Bond strength to permanent teeth affected by hypocalcified AI is lower than that to sound teeth.[24] In adults, bonded porcelain veneers were often used for anterior teeth in the hypoplastic type AI. However, complete-coverage crowns are the preferred restoration for patients with hypomature or hypocalcified type AI who experience severe sensitivity due to enamel structure loss.[25,26] Indeed, the risk of enamel breakdown is important for the palatal surface of maxillary anterior teeth, particularly if mandibular incisors are in functional contact. Moreover, in cases with severe discoloration, full-coverage restorations are required to properly restore teeth. Some authors have been described the use of all-ceramic crowns as a possible restorative approach particularly in the hypoplastic form.[27,28] Unfortunately, clinical follow-up is often short or unspecified. Although all-ceramic systems offer certain advantages (esthetics, biocompatibility, preserving hard tissues), chipping is reported to be a major complication associated with the use of all-ceramic materials, especially zirconia.[27,29-31] For anterior crowns, many all-ceramic restorations were found to demonstrate acceptable longevity compared with metal-ceramic restorations. However, for posterior tooth restoration, recent reviews suggest that relatively few all-ceramic systems will provide predictable long-term success.[29,31,32] In the case of our patient, duration of transitional, orthodontic, maxillofacial and prosthodontic treatment phases was considerable (period of 8 years). The patient wished a reliable rehabilitation to reduce the risk of reintervention. Considering these reasons and the difficulty in bonding hypomineralized enamel, the choice was made to use precious metal alloy with low-fusing ceramic for both high precision of execution and good long-term results. Indeed, these materials provide a lower hardness factor on the occlusal surfaces and may decrease the risk of fracture. We have treated other cases without complications after 10 years or more.[25,33,34] Urzua et al reported a case of hypocalcified AI rehabilitated with 28 ceramic metal-free

crowns cemented with adhesive techniques. Their treatments showed acceptable esthetic, biological and functional conditions after 4 years.[23] Even though the follow-up is not yet significant, the results obtained with all-ceramic lithium disilicate crowns are promising.[28,35] Future studies with long-term follow-up will be helpful in evaluating long-term results of all-ceramic restorations.

CONCLUSION

Full-mouth rehabilitation of patients with severe form of AI and AOB is a challenge for the dental team. A combination of orthodontic treatment, orthognathic surgery and prosthodontics rehabilitation are essential to successfully achieve esthetic and functional result.

CLINICAL SIGNIFICANCE

Complete restoration of severe AI is a long and complex process requiring a carefully established protocol for both initial treatment and long-term prosthetic phase, generally extending over several years. Early interdisciplinary treatment greatly enhances both dental esthetics and quality of life and also improves patient's self-confidence.

REFERENCES

1. Witkop CJ. Amelogenesis imperfecta, dentinogenesis imperfecta and dentin dysplasia revisited: Problems in classification. J Oral Pathol 1988;17:547-53.
2. Seow WK. Clinical diagnosis and management strategies of amelogenesis imperfecta variants. Pediatr Dent 1993;15: 384-93.
3. Sundell S. Hereditary amelogenesis imperfecta: An epidemiological, genetic and clinical study in a Swedish child population. Swed Dental J Suppl 1986;31:4-38.
4. American Academy of Pediatric Dentistry Council on Clinical Affairs: Guideline on oral health care/dental management of heritable dental development anomalies. Pediatr Dent 2008–2009;30(7Suppl):196-201.
5. Chan HC, Estrella NM, Milkovich RN, Kim JW, Simmer JP, Hu JC. Target gene analyses of 39 amelogenesis imperfecta kindreds. Eur J Oral Sci 2011;119(Suppl 1):311-23.
6. Wright JT, Torain M, Long K, Seow K, Crawford P, Aldred MJ, et al. Amelogenesis imperfecta: Genotype-phenotype studies in 71 families. Cells Tissues Organs 2011;194(2-4):279-83.
7. Witkop CJ, Sauk JJ. Heritable defects of enamel. In: Steward RE, Prescott GH (Eds). Oral facial genetics. St Louis: CV Mosby Co 1976:151-226.
8. Macedo GO, Tunes RS, Motta AC, Passador-Santos F, Grisi MM, Souza SL, et al. Amelogenesis imperfecta and unusual gingival hyperplasia. J Periodontol 2005;76(9):1563-66.
9. Thompson GA, Schwartz JM. Oral rehabilitation of a patient with amelogenesis imperfecta. J Prosthodont 1997;6:257-64.
10. Hoppenreijs TJ, Freihofer HP, Stoelinga PJ, Tuinzing DB, van't Hof MA, van der Linden FP, et al. Skeletal and dento-alveolar stability of Le Fort I intrusion osteotomies and bimaxillary osteotomies in anterior open bite deformities. A retrospective three-centre study. Int J Oral Maxillofac Surg 1997;26(3): 161-75.
11. Ravassipour DB, Powell CM, Phillips CL, Hart PS, Hart TC, Boyd C, et al. Variation in dental and skeletal open bite malocclusion in humans with amelogenesis imperfecta. Arch Oral Biol 2005;50(7):611-23.
12. 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;136:620-30.
13. Pires Dos Santos AP, Cabral CM, Moliterno LF, Oliveira BH. Amelogenesis imperfecta: Report of a successful transitional treatment in the mixed dentition. J Dent Child 2008;75(2): 201-06.
14. Varela Morales M, Botella Perez JM, Jiménez Garcia J, Varela PG. Interdisciplinary treatment of a patient with amelogenesis imperfecta. J Clin Orthod 2010;44(9):553-59.
15. Chan KH, Ho EH, Botelho MG, Pow EH. Rehabilitation of amelogenesis imperfecta using a reorganized approach: A case report. Quintessence Int 2011;42:385-91.
16. Doruk C, Ozturk F, Sari F, Turgut M. Restoring function and aesthetics in a Class II Division 1 patient with amelogenesis imperfecta: A clinical report. Eur J Dent 2011;5(2):220-28.
17. Wright J, Waite P, Mueninghoff L, Sarver DM. The multidisciplinary approach managing enamel defects. J Am Dent Assoc 1991;122:62-65.
18. Cartwright AR, Kula K, Wright TJ. Craniofacial features associated with amelogenesis imperfecta. J Craniofac Genet Dev Biol 1999;19(3):148-56.
19. Aren G, Ozdemir D, Sepet E, Firatli E. Evaluation of oral and systemic manifestations in an amelogenesis imperfecta population. J Dent 2003;31(8):585-91.
20. Ramos AL, Pascotto RC, Iwaki Filho L, Hayacibara RM, Boselli G. Interdisciplinary treatment for a patient with open-bite malocclusion and amelogenesis imperfecta. Am J Orthod Dentofacial Orthop 2011;139(4 Suppl):S145-53.
21. Hoppenreijs TJ, Voorsmit RA, Freihofer HP, van't Hof MA. Open bite deformity in amelogenesis imperfecta. Part 2: Le Fort I osteotomies and treatment results. J Craniomaxillofac Surg 1998;26(5):286-93.
22. Gjørup H, Haubek D, Hintze H, Haukali G, Løvschall H, Hertz JM, et al. Hypocalcified type of amelogenesis imperfecta in a large family: Clinical, radiographic, and histological findings, associated dento-facial anomalies, and resulting treatment load. Acta Odontol Scand 2009;18:1-8.
23. Urzúa B, Ortega-Pinto A, Farias DA, Franco E, Morales-Bozo I, Moncada G, et al. A multidisciplinary approach for the diagnosis of hypocalcified amelogenesis imperfecta in two Chilean families. Acta Odontol Scand 2012;70(1):7-14.
24. Faria-e-Silva AL, De Moraes RR, Menezes Mde S, Capanema RR, De Moura AS, Martelli H Jr. Hardness and microshear bond strength to enamel and dentin of permanent teeth with hypocalcified amelogenesis imperfecta. Int J Paediatr Dent 2011;21(4):314-20.
25. Bouvier D, Duprez JP, Pirel C, Vincent B. Amelogenesis imperfecta—a prosthetic rehabilitation: A clinical report. J Prosthet Dent 1999;82(2):130-31.
26. Kumar S, Gupta S. The restoration of function and esthetics of a patient with amelogenesis imperfecta using a combination of orthodontic and prosthodontic treatment: A case report. J Contemp Dent Pract 2009;10(6):e079-85.

27. Siadat H, Alikhasi M, Mirfazaelian A. Rehabilitation of a patient with amelogenesis imperfecta using all-ceramic crowns: A clinical report. J Prosthet Dent 2007;98(2):85-88.
28. Gisler V, Enkling N, Zix J, Kim K, Kellerhoff NM, Mericske-Stern R. A multidisciplinary approach to the functional and esthetic rehabilitation of amelogenesis imperfecta and open bite deformity: A case report. J Esthet Restor Dent 2010;22(5):282-93.
29. Della Bona A, Kelly JR. The clinical success of all-ceramic restorations. J Am Dent Assoc 2008;139(Suppl):8-13.
30. Groten M. Complex all-ceramic rehabilitation of a young patient with a severely compromised dentition: A case report. Quintessence Int 2009;40:19-27.
31. Land MF, Hopp CD. Survival rates of all-ceramic systems differ by clinical indication and fabrication method. J Evid Based Dent Pract 2010;10:37-38.
32. Rinke S, Tsigaras A, Huels A, Roediger M. An 18-year retrospective evaluation of glass-infiltrated alumina crowns. Quintessence Int 2011;42(8):625-33.
33. Bouvier D, Leheis B, Duprez JP, Bittar E, Coudert JL. Dentinogenesis imperfecta: Long-term rehabilitation in a child. J Dent Child 2008;75(2):192-96.
34. Millet C, Viennot S, Duprez JP. Case report: Rehabilitation of a child with dentinogenesis imperfecta and congenitally missing lateral incisors. Eur Arch Paediatr Dent 2010;11(5):256-60.
35. Edelhoff D, Brix O. All-ceramic restorations in different indications: A case series. J Am Dent Assoc 2011;142 (Suppl 2):14S-19S.

ABOUT THE AUTHORS

Catherine Millet (Corresponding Author)

Professor, Department of Prosthodontics, Faculty of Dentistry University of Lyon, F-69622; Centre for Oral Manifestations of Rare Diseases, Hospices Civils de Lyon, Lyon France, e-mail: cathymillet@yahoo.fr

Jean-Pierre Duprez

Assistant Professor, Department of Pediatric Dentistry, Faculty of Dentistry, University of Lyon, F-69622; Centre for Oral Manifestations of Rare Diseases, Hospices Civils de Lyon, Lyon France