

Oral rehabilitation of a patient with amelogenesis imperfecta - a case report

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Abstract

Amelogenesis imperfect (AI) is a hereditary condition where enamel formation is disturbed resulting in defects in mineralization or matrix formation. Restoration of the dentition poses great difficulties especially when all the teeth are severely affected. Treatment aims to relieve pain or tooth sensitivity, to preserve as much tooth tissue as possible while preventing further tooth loss, to maintain masticatory function, and, last but not least, to improve the appearance as this has great psychological impact on the patient's confidence. This article discusses both the functional and esthetic rehabilitation of a patient with hypoplastic type of AI. Both the esthetics and function were hampered in this patient due to the condition. As a result, the treatment was properly planned and executed.

Key words: Amelogenesis imperfecta (AI), rehabilitation, hypoplastic type, esthetics.

Introduction

Amelogenesis imperfecta (AI) is a diverse collection of inherent disease that exhibit quantitative or qualitative enamel defect in the absence of systemic complication. It is a hereditary disorder that expresses a group of conditions that cause developmental alterations in the structure of enamel. The incidence of AI has been reported as varying between 1:700 and 1:16000, depending on the diagnostic criteria and the population studied⁽¹⁾. The estimated frequency of AI in the population varies between 1:718 and 1:14.000 in western population. Generally both the primary and permanent dentitions are diffusely involved⁽²⁾. In the affected teeth, the dentin and root form are usually normal. No racial predilections of the AI have been reported.

Classifications of AI are primarily based on phenotype and mode of inheritance. The most commonly used classification was proposed in 1988 by Witkop, and revised by Nusier in 2004⁽¹⁾. Based on enamel appearance and hypothesized developmental defects, AI is classified as four patterns: hypoplastic, hypomaturation, hypocalcified, and hypomaturation-hypoplastic^(1,3). The primary clinical problems of AI are tooth sensitivity, loss of occlusal vertical dimension, dysfunction, and esthetics. Restoration of these defects is important not only because of esthetic and functional concerns, but also because there may be a positive psychological impact for the patient. The trait of AI can be transmitted by an autosomal dominant, autosomal recessive, or X-linked mode of inheritance^(4,5). The distribution of AI types is known to vary among different

populations. In a study in Sweden, 63% of the cases were inherited as autosomal dominant. In contrast, in a study in the Middle East, the most common prevalent type of AI was found to be autosomal recessive^(6,7).

Treatment planning for patients with AI is related to many factors; the age and socioeconomic status of the patient, the type and severity of the disorder, and the intraoral situation. An interdisciplinary approach is necessary to evaluate, diagnose, and resolve esthetic problems using a combination of periodontal, prosthodontic, and restorative treatment. The aim of this paper is to outline the management of esthetics with periodontal approach in a patient with AI of the hypoplastic type.

Case Report

An 18 year old girl reported to the outpatient Department of Periodontics, A.J institute of Dental Sciences, Mangalore, Karnataka, India with a chief complaint of yellow discoloration and poor appearance of her teeth. Her medical history was non contributory. Her past dental history revealed extraction of 26 due to caries and also no report of any parafunctional habits. No other family member had the same dental problem. Extraoral examination did not reveal any relevant findings. The patient's oral hygiene was poor and presented hyperaemic and oedematous gingiva. Gingival enlargement was seen in most of the anterior teeth and the clinical crown length was inadequate. Periodontal pocket of 8mm was present in relation to 46.

Hard tissue examination revealed abnormally small sized



Figure 1. Clinical picture showing deep pitting and discoloration of the teeth

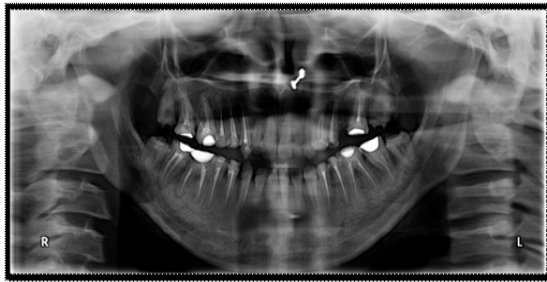


Figure 2. Radiograph prior to periodontal treatment



Figure 3. Maxillary and mandibular casts obtained before periodontal surgery

teeth, the enamel was thin on most of the areas and absent on the occlusal surfaces due to attrition, and surfaces of the teeth were rough with calculus deposits. Teeth revealed a defined and deep pitting with discoloration (**Figure 1**). The thickness of enamel was reduced and partially chipped off from few of the teeth surfaces. Dentine which was exposed was brown in color. Chronic peri-radicular periodontitis (chronic apical periodontitis) was present in relation to 35, 37 and 47.

Radiographic examination revealed maxillary and mandibular third molars and a seemingly normal pattern and timing of eruption of teeth. The pulp chamber was

normal and root canal spaces with no signs of obliteration. Presence of periapical radiolucency with widening of periodontal ligament space was seen in relation to 35, 37 and 47. Radiograph also showed the presence of bone loss in relation to 46. The enamel layers were absent in all the teeth giving an appearance similar to that observed after mechanical preparation (**Figure 2**).

Complete maxillary and mandibular impressions were made and diagnostic casts were obtained (**Figure 3**). Pre-operative intraoral photographs and radiographs were also obtained. Once these were done, complete treatment plan was explained to the patient. All factors including the amount of tooth structure removal, soft tissue surgery, need for endodontic therapy and duration of the treatment were discussed with the patient and written informed consent was obtained.

Initial stage of treatment consisted of oral hygiene instructions, scaling and root planning. Two weeks later the gingival edema was resolved and there was no bleeding on probing. After the initial periodontal treatment, pulp space therapy (root canal treatment) in relation to 13, 14, 16, 17, 24, 25, 27, 34, 35, 36, 37, 44, 45, 46 and 47 was performed.

The patient required crown restoration for aesthetic and functional purpose. The patient had inadequate tooth structure for placement and retention of restoration taking into consideration the attachment apparatus. Hence the main objective of the surgery was to provide adequate tooth structure for the placement of crown restoration.

Determination of the desired gingival margin and bony crest were completed pre-operatively to ensure the success of crown lengthening. Factors for evaluation included: general characteristics of patient, the proximity of surrounding anatomic structure, gingival thickness, biologic width, gingival recession, and quality of remaining tooth structure and furcation location. Periodontal flap surgery was carried out for four quadrants. An internal bevel incision was designed in buccal and palatal area to contour the soft tissues. Full thickness mucoperiosteal flap was raised, and osteotomy procedure was designed to achieve at least 3mm of supracrestal sound tooth structure within the interdental area in relation to 16, 17 and 27 (**Figures 4 & 5**). Osteoplasty was performed in relation to 14, 15, 24, 25,



Figure 4. Mucoperiosteal flap reflection prior to osseous correction



Figure 5. Osteoplasty procedure to obtain positive architecture



Figure 6. Post operative photograph following crown lengthening with osseous correction



Figure 7. Clinical photograph of patient with final prosthetic restoration

34,35,36,37,44,45,46 and 47 in order to achieve positive architecture. In relation to 46 intrabony defect was treated with bone graft material. Buccal and palatal flap was sutured with interrupted sutures. Periodontal pack was placed. The surgery was done in such a way that there was no compromise regarding the biologic width. The surgical procedure was completed in a span of 4 weeks.

After the completion of the surgery post surgical instructions were given and oral hygiene reinforced. We waited for 3-4 weeks for healing to take place after which the patient was referred to the Department of Prosthodontics for metal ceramic crowns. Follow up visits

were scheduled at 3 and 6 months. No esthetic or functional problems were seen during the follow up period (Figures 6 & 7).

Discussion

One of the most challenging dilemmas for a clinician is the treatment of a teenage patient with multiple defects of the enamel like AI and its associated conditions. The affected enamel is disturbed in coloration, thickness and resistance. As a result these patients are more prone to tooth decay, periodontal disease and occlusal disturbances. In this manner AI is a condition that seriously compromises the oral and psychological health of the patient and it requires early recognition and action. Nearly all patients affected with AI assessed themselves to be aesthetically disturbed by their pretreatment condition. The condition negatively affected the relationship with other people and their self esteem.

There are a number of treatment options for teeth affected by AI. The treatment of patients with AI presents a challenge to the clinician. The treatment plan usually varies and many factors have to be taken into consideration age of the patient, socioeconomic status, and severity of the disorder. The need for crown lengthening is dictated by dental and patient factors. Crown lengthening may be as simple as a limited removal of soft tissue or as complex as orthodontic extrusion followed by flap with osseous surgery on a tooth requiring endodontic therapy. Total treatment could thus involve endodontic, orthodontic, periodontic, and restorative procedures. Careful evaluation, case selection, treatment planning, and surgical treatment following the principles can achieve results that meet the functional and esthetic challenges of current dental practice⁽⁸⁾. Although for crown lengthening gingivectomy (external bevel incision) is the choice of treatment, in our case report a full thickness mucoperiosteal flap was raised, and osteotomy procedure was designed to achieve at least 3mm of supracrestal sound tooth structure and osteoplasty was performed in order to achieve positive architecture.

A study was conducted by Wayne et al. in 2000⁽⁹⁾ treated a patient with AI with two surgical procedures. In the first surgical procedure full mouth flap surgery for all four quadrants was carried out to allow the soft tissue to mature and the second surgical procedure was carried out to correct the soft tissue symmetry and form. Contrary to this study our study aimed at soft and hard tissue contouring and achieved positive results without the patient undergoing two surgical procedures.

After crown lengthening it should be possible to put restoration margins either coronal, or at, the gingival margin. It is well documented in the literature that this creates a more favorable condition to allow periodontal health. It was found that margins of fixed prosthodontics significantly compromise the gingival health, if placed below the gingival margin⁽¹⁰⁾. In a study it was found that subgingival margins demonstrated higher plaque, gingival index scores, and probing depths. In addition, when the bacterial morphotypes were examined, there was an increase in the spirochetes, fusiforms, rods, and filamentous bacteria⁽¹¹⁾. Hence, periodontal health is the cornerstone of any successful restorative procedure. Therefore, the correct handling of the periodontal tissues

during restoration of the tooth is important to the restoration's future success⁽¹²⁾.

This case report described the oral rehabilitation of a young adult patient affected by hypoplastic AI. Although the combination of Orthognathic surgery and orthodontic treatment was explained to the patient as part of the optimum treatment plan to achieve the best occlusal result, the patient declined this option due to the significant financial burden and surgical procedure involved.

Conclusion

In conclusion, AI is a serious problem that can result in reduced oral health-related quality of life and causes some physiological problems. From this point of view, people with AI need extensive treatment. While planning the treatment, the age and the socioeconomic status of the patient, type and the severity of the disorder should be taken into consideration. This multidisciplinary treatment plan should be considered for indicated situation while implementing careful evaluation, case selection, treatment planning and precise surgical and associated treatment in order to meet the functional and aesthetic demands of current dentistry.

References

1. Witkop CJ Jr. Amelogenesis imperfecta, dentinogenesis imperfect and dentin dysplasias revisited. Problems in classification. *J Oral Pathol* 1988; 17:547-553.
2. Aldred MJ, Crewford PJ. Amelogenesis imperfecta: Towards a new classification. *Oral Dis* 1995; 1:2-5.
3. Backman B, Holmgren G. Amelogenesis imperfecta: A genetic study. *Hum Hered* 1988; 38:189-206.
4. Aldred MJ, Savarirayan R, Crawford PJ. Amelogenesis imperfecta: A classification and catalogue for the 21st century. *Oral Dis* 2003; 9:19-23.
5. Backman B. Inherited enamel defects. *Ciba Found Symp* 1997; 205:175-182.
6. Chosack A, Eindelman E, Wisotski I, Cohen T. Amelogenesis imperfecta among Israeli jews and the description of a new type of local hypoplastic autosomal recessive amelogenesis imperfecta. *Oral Surg Oral Med Oral Pathol* 1979; 47:148-156.
7. Nusier M, Yassin O, Hart TC, Samini A, Wright JT. Phenotypic diversity and revision of the nomenclature for autosomal recessive amelogenesis imperfecta. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2004; 97:220-230.
8. Allen EP. Surgical crown lengthening for function and esthetics. *Dent Clin North* 1993; 37:163-179.
9. Williams WP, Becker LH. Amelogenesis imperfecta: functional and esthetic restoration of a severely compromised dentition. *Quintessence Int* 2000; 31:397-403.
10. Silness J. Fixed prosthodontics and periodontal health. *Dent Clin North Am* 1980; 24:317-339.
11. Flores-de-Jacoby L, Zafiropoulos GG, Ciancio S. The effect of crown margin location on plaque and periodontal health. *Int J Periodontics Restorative Dent* 1989; 9:197-205.
12. Gargiulo AW, Wentz F, Orban B. Dimensions and relations of the dentogingival junction in humans. *J Periodontol* 1961; 32:261-267.