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Indirect adhesive rehabilitation by cementation under pressure of a case of Dentinogenesis Imperfecta type II: follow-up after 13 years

ABSTRACT

Background *Dentinogenesis imperfecta (DI) is an autosomal dominant genetic disease that affects both deciduous and permanent teeth, with an incidence of 1 out of 6,000 to 1 out of 8,000. Teeth affected with DI type II present bulbous crowns, short and constricted roots, marked cervical constriction, translucent enamel and amber dentin. Also, they present a partial or total obliteration of pulp space, due to continuous dentin production. SEM analysis has shown an undulated dentin-enamel junction (DEJ) with irregularities and locally wide spaces between the two structures instead of a strict junction and a regular linear surface. Treatment options for patients affected by DI-II are intended to protect and restore function and aesthetics of both posterior and anterior teeth. In literature are presented many different therapies, but mainly centered on cemented prosthetic restorations instead of adhesive restorative procedures. We present in this paper a DI-II case successfully treated in 2005 with extensive adhesive rehabilitation.*

Conclusion *The 13 years follow-up proves the reliability of adhesion to dentine and enamel for indirect adhesive restorations even on this kind of anomalous substrates.*

Keywords *Dentinogenesis Imperfecta, Adhesive Restorative rehabilitation.*

Introduction

Dentinogenesis Imperfecta (DI) is an autosomal dominant genetic disease with an incidence of 1 out of 6,000 to 1 out of 8,000 [Witkop, 1957]. This disorder has been classified into three subtypes: type I (DI-I) also called "syndromic" and associated with Osteogenesis Imperfecta (OI) types III and IV; type II (DI-II), "non-syndromic" and limited to dentin; and type III (DI-III) or "hereditary opalescent dentin" (Brandywine), initially detected in southern Maryland (USA) [Shields et al., 1973] and characterised by larger pulp chambers that lead to multiple pulp exposure mainly in deciduous dentition [Hart and Hart, 2007].

DI-II is due to mutations in the DSPP gene located at 4q22. DSPP is responsible for producing two principal non-collagenous proteins of mature dentin matrix (DPP and DSP) that may be involved in the nucleation and modulation of apatite crystal formation, and thus in the proper shape and placement of the crystals in dentin matrix [Wang et al., 2011; Wieczorek, 2013; Kawashima, 2016]. Clinically DI-II is characterised by opalescent and amber dentin and thin, translucent and greyish-brown enamel. Radiographically DI-II affected teeth have a bulbous crown, short and constricted roots and partial or total obliteration of pulp space, due to continuous dentin production [Gallusi et al., 2006].

SEM analysis has shown an undulated dentin-enamel junction (DEJ) with irregularities and locally wide spaces between the two structures instead of a strict junction and a regular linear surface [Gallusi et al., 2006].

Other studies [Wieczorek and Loster, 2013] highlighted typical features of DI-II enamel and dentin. The enamel surface is irregular because the hydroxyapatite crystals in the enamel prisms spread in different directions, plainly showing a weakened tissue structure. Dentin is both atubular or with few wide tubules, and collagen fibers spread in different directions along the walls.

Since the enamel-defective structure is confined to the occlusal third of the crown, enamel defects may not only be due to attrition or fracture, but to a negative influence of abnormal DSPP gene products on ameloblast and dentinoblast functions [Lee et al., 2011].

Treatment of DI-II has been widely discussed in scientific literature [Sapir et al., 2001; Sapir et al., 2007; Moini et al., 2013; Moreira et al., 2015; Akhlagi et al., 2016]. Treatment modalities mainly consisted in prosthetic rehabilitation of posterior teeth (second deciduous molars and first and second permanent molars) through placement of overdentures, stainless steel crown, jacket crowns, pin-retained cast gold thimbles under acrylic resin crowns, stainless steel crowns with acrylic facing, simple removable appliances, indirect resin crown to guarantee protection and function of the crowned teeth.

Moreover, in some follow-ups reported over the years, it was clearly shown how the teeth treated in this way had cervical/gingival lesions and fractures due to the

difficulty in keeping a good level of oral hygiene at home, resulting in bone loss and need of extractions [Akhlagi et al., 2016].

Case report

The patient in this paper had been visited for the first time at the age of 7 years. The approach that was used in such a young patient was to instruct him to have proper oral hygiene and to seal pits and fissures, in agreement with a study conducted in 2005 by Albani et al., in which a 75% decrease in the incidence of occlusal decay was demonstrated in first and second permanent molar previously treated with sealing pits and fissures.

Once reached adolescence (17 years old), a full and multidisciplinary rehabilitation was finally possible. After 10 years from sealer placement, this kind of preventive approach has been very successful. At this time there are no clinical and radiographic signs of caries (Fig. 1). The main problem for the patient was the large loss of dental tissue (caused by enamel abrasion and fracture), which obviously resulted in a loss of vertical dimension limiting function and aesthetics.

In the literature, patients with enamel and dentine defects were mainly treated according to a prosthetic approach. Patients affected by DI-II unfortunately have dental structure and also teeth shape anomalies, making prosthetic procedures not the best choice. The height of teeth crowns is often so low that a crown lengthening procedure would be necessary prior to prosthetic preparations. At the same time, shortness of roots and the bulbous shape of DI-II teeth contraindicate any crown lengthening procedure. For this reason, the authors decided to adopt a restorative approach rather than a prosthetic one, in order to preserve as much enamel as possible with full indirect adhesive restorations.

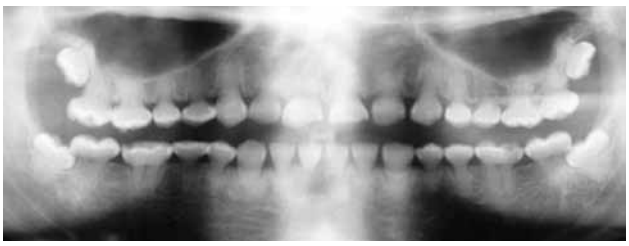

FIG. 1 Panoramic X-ray of a patient affected with DI-II in 2005 (pre-treatment). Typical teeth aspect with bulbous crown, short and constricted roots and partial obliteration of pulp space.

The posterior teeth were restored with indirect composite resin overlays cemented with the same light curing composite resin. The chosen adhesive cementation strategy follows the concept of "cementation under pressure" of indirect adhesive restorations: rubber dam insulation, 37% phosphoric acid etching, adhesion to dentin and enamel with a two-step light curing bonding [Libonati et al., 2011; Ballesio et al., 2012; D'Amario et al., 2015], and finally cementation under pressure with light curing of the indirect composite resin overlays [Campanella, 2003] (Fig. 2).

Due to the abnormal dental substrates present in the DI-II, it is expected to achieve a less than ideal hybrid layer as a result of even the best dental bonding procedures. For this reason, the authors chose indirect resin restorations with all margins in enamel, highest possible precision, perfect marginal adaptation and cementation under pressure with highly charged light curing micro-hybrid composite resin [Campanella, 2003; Gallusi et al., 2009]. The "cementation under pressure" technique consists of applying high pressure to the indirect restoration fully loaded with 40 °C preheated light curing composite while seating it on the tooth. Achieved the complete seating of the overlay, the release of the pressure would cause the restoration to move back as a result of the pressure built inside of it; this is the moment for increasing the pressure over the restoration, remove grossly the excess of cementing material and light cure the composite resin for an extended period of time using high power light curing units. Very precise indirect composite restorations are also a good choice to deal with polymerisation contraction of composite resins. The amount of composite resin to be cured during cementation is very low and put under pressure during conversion leading to low shrinkage and better mechanical property. These restorations are very stable, as a chemical and micromechanical bond is formed between the composite used as cement, the inlay (already polymerised in the laboratory) and prepared dentine and enamel. This restorative strategy can reduce

FIG. 2 Indirect composite restorations in posterior teeth immediately after cementation in 2005.

FIG. 3 Ceramic veneers (Empress) on anterior teeth in 2005.

internal stresses preserving the marginal enamel from undesired micro-cracks thus achieving a better marginal seal.

The anterior teeth were prepared for Empress ceramic veneers. The preparation involved the vestibular and proximal surfaces wit palatal but-joints and vestibular chamfer margins.

For the luting procedures, after a careful rubber dam insulation of all prepared teeth, each veneer was etched with 9.5% hydrofluoric acid, treated with silane and bonded with fluid resin. The veneers were cemented with the same 40 °C preheated light curing microhybrid composite resin used for posterior restorations. During the cementation of the veneers a little portion of the enamel on a margin failed and separated probably due to the abnormal histology of DEJ in DI-II and the stress induced by dehydration (this was discussed on the original case report published in 2006). The cementation technique we used was able to resolve the problem, since every inconsistency between ceramic and the prepared toot was restored and sealed with light curing microhybrid composite used in perfectly controlled and insulated field (Fig. 3).

The 13-year follow-up confirmed the possibility to successfully treat these cases with less invasive adhesive restorations. Accurate and precise preparations, high precision indirect restorations and rigorous under

FIG. 4 At the 7-year follow-up (2012) the anterior restorations show a loss of brilliance due to the aging of the veneers, with minimal gingival recession in some areas.

pressure cementation procedures were able to restore function and aesthetics and preserve the obtained result for a long period of time with great satisfaction of the patient (Fig. 4-8).

The radiographic examination shows the long-lasting periodontal health of the rehabilitated teeth due to the correct prosthetic profile maintained over time (Fig. 9) [Concolino et al., 2007; Tecco et al., 2011; Germano et al., 2013; Mummolo et al., 2014; Tundo et al., 2015].

This rehabilitation did not present, after 13 years of use, any issue or adhesive failure. Comparing the present appearance of the restoration with initial photos it is clear that there is a kind of aging but with no adverse effects. Due to the kind of restorations, if it will be necessary in case of local failures, it will be always possible to perform

FIG. 5 At the 7-year follow-up (2012) the posterior restorations show a good performance and integrity of the composites..

FIG. 6 Panoramic X-ray 7 years after treatment (2012). FIG. 7 The 13-years follow-up (2018) showed an exposure of the margins in some areas without any functional implication and having an acceptable aesthetic compromise.

FIG. 8 The 13-years follow-up (2018) showed a distal and palatal fracture of the element 2.5; a loss of the top of the distal-vestibular cusp of the element 2.6; a slight loss of the distal marginal crest of the element 3.6; an occlusal cavity of the element 3.7; and a lingual crack of tooth 4.6.

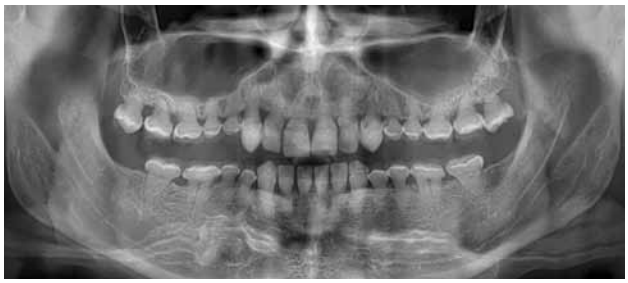

FIG. 9 Panoramic X-ray 13 years after-treatment (2018), showing the radiopacity of the anterior ceramic veneers and the radiolucency of the cemented posterior restorations. No carious process is underway. There is also a diffuse horizontal bone defect.

FIG. 10 Intraoral photographs of superior palatal view, 2018. Fractures with no detachment of the ceramic in the palatal area of the elements 2.1 and 2.2 are highlighted.

FIG. 11 Intraoral photographs of inferior lingual view, 2018. Although the patient does not have good level of oral hygiene, the integrity of the restorations until today has been maintained.

new restorations or adhesive repairs preserving all the remaining sound tissues (Fig. 10, 11).

The long-term result confirms the choice of a restorative treatment plan. The success in complex cases like this is always due to a multidisciplinary approach and highly rigorous procedures that are necessary for a predictable clinical success.

Acknowledgements

the Authors declare that they are free of conflicts of interest.

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