Soliman et al.  

Treatment of an Adolescent Patient with Dentinogenesis Imperfecta Using Indirect Composite Restorations – A Case Report and Literature Review
Purpose: To demonstrate the field of application and prospects of individually modeled indirect composite restorations for the treatment of children and adolescents based on a case of dentinogenesis imperfecta. Dental malformations can affect single or multiple teeth. In most cases, direct composite fillings can be placed. However, in severe cases, these restorations may be more challenging and error-prone, especially when occlusal adjustments are necessary. Since composite materials do not require a specific lamination strength and are easy to repair, they can be applied using the indirect technique, enabling conservation of more sound hard tissue than is possible when conventional restorations are used.

Patient and Methods: A young patient with dentinogenesis imperfecta type II underwent interdisciplinary full-mouth rehabilitation due to massive tooth wear and loss of vertical occlusion. First, a check bite was taken, and vertical occlusion was increased using overdentures. Six months later, a construction bite was taken over the existing overdentures (focusing on the sagittal dimension) to move the mandibular position more towards the anterior, correcting the skeletal Class II malocclusion. This resulted in a Class I intercuspidation with harmonization of the facial proportions. After a further six months, all teeth were restored using individually modeled indirect composite restorations, which preserved most of the sound hard tissue and restored esthetics and function.

Conclusion: Indirect composite restorations can be a valuable tool for improving occlusion, esthetics and function in the treatment of children and adolescents.

Keywords: severe tooth wear, dental malformations, full-mouth rehabilitation, individual lamination technique, fiber reinforced composite restorations, overdenture, hereditary disorder.

REVIEW OF THE LITERATURE

Pathogenesis of Dentinogenesis Imperfecta (DI)

DI is a hereditary disorder that shows autosomal dominant inheritance. Three types of DI have been described by...
Shields et al. Type I is associated with osteogenesis imperfecta and originates from a collagen-I defect. The degree of dental malformation can vary across the primary and the permanent dentition, and in some cases the teeth have only radiographic abnormalities but no clinical damage. In contrast, DI type II shows severe dental malformations in both the primary and the permanent dentition and full penetration within a given family. The associated malformations are related to a mutation of the dentin sialophosphoprotein gene, which is located on chromosome 4q21. DI type III was primarily found in a particular ethnic group in the USA and described by Witkop et al. and Hursey et al. The deciduous teeth of type III can have a shell-like shape with very short or even missing roots, whereas the permanent teeth resemble those found in the other types of DI. Other genetic studies have revealed that Shields type II and III might be allelic and may represent different severities of the same disorder. Furthermore, these studies suggest that dentin dysplasia type II is also a phenotype of this DI.

Whereas DI type I has a greater range of clinical severity, DI types II and III are consistently characterized by amber-colored dentin, opalescent and bulbous crowns and constrictions at the cementoenamel junction (CEJ). Observations from radiographs reveal that in addition to the typically shortened and narrowed roots, the pulp chamber is quickly calcified when tooth eruption begins. Histological examinations of the dental hard tissue of these patients reveal regularly structured enamel but aberrations in the structure of the dentin and the enamel-dentin junction. Generally, the number and size of the dentinal tubules are reduced, and the apatite crystallites have an abnormal morphology, while the water and collagen content is increased. The adherence between the enamel and dentin is compromised and the enamel chips off, resulting in severe abrasion and attrition. Lindau et al. ascribed this poor adherence to a malfunction in the interaction between the ectodermal and mesenchymal tissues. This rapidly leads to a loss of vertical occlusion even shortly after tooth eruption begins. Consequently, craniofacial dysgnathia, esthetic deficiencies, and disorders in function and articulation may occur. In many cases, these patients do not consult specialists until esthetics are massively compromised or symptoms arise, at which point they show extensive tooth destruction.

**Therapy Regimes in DI**

Numerous case reports exist describing different treatment procedures, most of which involve the prosthetic treatment of missing teeth. Few present early conservative non-destructive therapies that can preserve these teeth sustainably. Preventing the early loss of teeth should be the goal of first interventions, as this simplifies later therapies tremendously and avoids the risk of impairing jaw growth and the loss of alveolar bone. This requires continuous visits and timely interventions, which can be slowed by the poor compliancy of children. Depending on the patient’s stage of life, different therapies are indicated.

For the primary dentition, stainless steel crowns and composite buildups are recommended for patients with enough residual hard tissue. In cases of severe loss of dental hard tissue, overdentures can be made, although they may hamper oral hygiene and may require further adjustments over time. The mixed dentition stage includes the most important and difficult part of therapy, as even partly erupted teeth begin to wear when they attain occlusion. Ideally, these teeth should be sealed with composite. Keeping these teeth dry during treatment can be difficult, possibly reducing the longevity of the sealants. Continuous follow-up visits are necessary to extend sealants or repair defective ones as tooth eruption proceeds. When the CEJ is accessible, the tooth should be covered completely, preferably with indirect restorations, because they ensure proper occlusal adjustment by the dental technician, unlike direct composite buildups. As in the present case, ICRs can here be considered a preservative and esthetically valuable option. When applicable, all teeth can be restored consecutively in this way, and repairs can remain functional past adolescence. Additionally, orthodontic treatments are easier to perform when the fragile enamel is covered. At this point, stainless steel crowns must be mentioned as an inexpensive alternative for the permanent posterior teeth; however, they have drawbacks in terms of esthetics and fit compared with ICRs. Additionally, once a stainless-steel crown is applied to a permanent tooth, any future restoration must be a full crown, whereas the use of ICRs leaves other options open.

In later adolescence or adulthood, however, when craniofacial growth is complete and the gingiva retracts, the margins of restorations may become visible and further full-mouth rehabilitation might be necessary to correct unacceptable esthetics. In the treatment of adults, the literature shows cases of full-mouth rehabilitation using all-ceramic restorations or full-mouth extractions and implant-supported dentures, depending on the individual’s dental chart.

The present article focusses on the treatment of the permanent dentition in adolescents by describing the case of a young patient with little experience of dental treatment. At this age, treatment must be carefully planned to consider the loss of dental hard tissue relative to longevity and esthetics. ICRs seem to be ideal in such cases for two reasons. First, thanks to adhesive bonding, no mechanical retention is necessary, which permits the restoration of very short teeth. In addition, these restorations can be made with a thickness of only 0.2 mm, so that in many cases, tooth preparation can be omitted or less invasive. In contrast, commonly used restorations (ie, metal-ceramic or all-ceramic) require sufficient hard tissue and tooth preparation. However, ICRs must be inserted under absolutely dry conditions, which cannot always be guaranteed. Furthermore, composites are still considered provisional, because they ensure proper occlusal adjustment by the dental technician, unlike direct composite buildups. As in the present case, ICRs can here be considered a preservative and esthetically valuable option. When applicable, all teeth can be restored consecutively in this way, and repairs can remain functional past adolescence. Additionally, orthodontic treatments are easier to perform when the fragile enamel is covered. At this point, stainless steel crowns must be mentioned as an inexpensive alternative for the permanent posterior teeth; however, they have drawbacks in terms of esthetics and fit compared with ICRs. Additionally, once a stainless-steel crown is applied to a permanent tooth, any future restoration must be a full crown, whereas the use of ICRs leaves other options open.

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restorations. This statement might not fully apply to patients with dental malformations; for instance, Incici et al.\textsuperscript{24} showed that the longevity of any kind of restoration is compromised in these patients. Nevertheless, a case series by Feierabend et al.\textsuperscript{14} revealed satisfactory results with ICRs.

**CASE REPORT**

A 10-year-old girl was referred by her dentist to the Department of Orthodontics at the University Hospital of Würzburg due to an Angle Class II malocclusion. The primary disease was diagnosed as DI type II. Severe tooth wear had caused a loss of vertical dimension and insufficient occlusion. The vertical loss was accompanied by skeletal Class II malocclusion, Angle Class II occlusion, and an increased overjet (8.0 mm). Extraorally, a negative lip profile was noticeable (Fig 1). When the patient was 10 years old, functional orthodontic therapy with a Class-II bionator was attempted to stimulate the growth of the mandible. At the same time, the patient’s former dentist covered the exposed dentin with composite sealants to reduce further tooth wear. Two years later, when the patient was 12 years old, orthodontic therapy was cancelled because of the patient’s poor compliance. Furthermore, the severe loss of dental hard tissue and the extremely short crowns made it impossible to attach orthodontic brackets. She was then referred to the Department of Operative Dentistry of the University Hospital of Würzburg for restorative therapy. At this point, the clinical findings revealed a negative smile line (Fig 2), reduced vertical bite height (Figs 3 to 5) with severely worn and decayed incisors, and completely worn first molars (Figs 6 and 7). The radiographic findings revealed no periapical lesions but presented the typical narrow, short roots with calcified root canals and bulb-shaped crowns (Fig 8).

**Pretreatment**

Planning and therapy were performed in collaboration with the orthodontist. First, two plaster models of each jaw were made and a check bite was performed using wax. The first pair of the models was mounted in an articulator (Protar 7
Articulator, KaVo; Biberach, Germany) using a face bow with the check bite in maximal intercuspidation (Fig 9). The intermaxillary space, intercuspation, and required space for the prospective restorations were analyzed and the future lengths of the incisors were estimated. Subsequently, we lowered the incisal pin of the articulator by 1 mm to achieve an increase in the vertical intermaxillary distance of 2 mm at the molars and 4 mm at the incisors. A diagnostic wax-up was performed on the casts (Fig 10) and the wax-up was cast with silicone to produce a mold for overdenture prostheses (Fig 11). The second pair of plaster models was mounted in the articulator in the same manner used for the first models, and thermoformable splints (Erkodur 1,0 mm hard, Erkodent; Pfalzgrafenweiler, Germany) were adapted and cut back to half of each occlusal tooth surface. Using the silicone molds, composite (Biodent, DeguDent; Hanau, Germany) was polymerized on the thermoformable splints to produce the overdenture prostheses (Fig 12). The fit of the overdentures and the length of the incisors were checked. No adhesive paste was needed to fix the overdentures, since their buccal margins were extended to the undercut areas of the natural teeth, providing sufficient friction.

Initially, the patient complained of myofacial pain in the temporal muscle, which was alleviated with physical therapy, speech therapy, and occlusal adjustments of the overdentures at the follow-up visits. The patient received instruction in oral hygiene and was advised to wear the overdentures for at least 16 h a day but to leave them out while eating to prevent fractures. The incisor length was adjusted during return visits to improve esthetics and pronunciation. Overall, the first phase of treatment took six months until the patient felt comfortable with her bite and pronunciation.

The second phase of treatment started with another analysis of intercuspidation and the proportion of the skull since the patient’s growth proceeded. The clinical findings revealed a slightly shortened lower face, a persistent negative lip profile (Fig 13), and a persistent Angle Class II malocclusion with distal occlusion of half the premolar size. To improve the intercuspidation and the facial proportions, we performed another silicone check bite (Imprint Bite, 3M Oral Care; St Paul, MN, USA) of the overdentures: A construction bite was made with the mandible in a forward position (by a half-premolar size) resulting in a bilateral Class I occlusion. Figure 14 shows balanced facial proportions of the patient with inserted overdentures and the new construction bite.

A face-bow was then applied over the maxillary overdenture and both jaws were then cast with alginate along with inserted overdentures. New plaster models were made jointly with the overdentures and mounted in the articulator using the new bite registration. To minimize
effort, the overdentures were then remodeled according to the design of an orthopedic twinblock appliance. Tilts were polymerized on each of them to inhibit retrusion of the mandible (Fig 15). Speech therapy and physical therapy were continued until the patient felt comfortable with the new maxillomandibular relationship, and the restorative stage began six months later. This long second waiting period was due more to the time-consuming correspondence with the patient’s medical insurance, which initially refused to assume therapy costs, rather than to medical reasons.

**Treatment – Restorative Stage**
First, the incisors and premolars were restored. To simplify the checkbite procedure after the prospective tooth preparation, a resin splint (Biodent, Dentsply; York, PA, USA) was created using new plaster models, which were mounted in the articulator in the same manner used in the overdenture remodeling procedure described above. The maxillary model was mounted using a face-bow appliance, the mandible model was mounted against it, and the overdentures were placed on both models. These were joined together by a silicone check bite before they were mounted (Fig 15), allowing the current intraoral relationship of the bite to be transferred to the future resin splint (Fig 16). The splint with...
Soliman et al

Fig 17 Pre-preparation of the incisors with inserted and silica-coated parapulpal retentive pins. Enamel of the lateral incisors was preserved since only undercuts were removed.

Fig 18 Composite buildups and chamfer preparation of the central incisors. Lateral incisors with removed undercuts and residual enamel.

Fig 19 Tooth preparation of the mandibular incisors and premolars showing removed undercuts with residual enamel and intact interproximal contacts.

Fig 20 Fine-impression by means of the resin splint and provisional cement. The splint was cut back at the maxillary incisor region since buildups were made.

Fig 21 The maxillary overdenture was cut back and fixed with adhesive paste. The incisors received conventional provisional crowns.

Fig 22 Completed individually modeled indirect composite restorations.

its impressions provided guidance for later registration after the tooth preparation was completed.

As all premolars and the mandibular incisors showed sufficient amounts of residual enamel (Figs 6 and 7), these teeth were prepared just by removing undercut areas and smoothing sharp edges to conserve as much enamel as possible. However, the maxillary incisors showed a strong degree of destruction, having no clinical crown height and only
a small ledge of enamel near the CEJ. Thus, composite build-ups were created and additionally retained by parapulpal retentive pins (TMS Link Plus, Self-threading pins, Coltene/Whaledent: Altstätten, Switzerland) (Fig 17). A previously made periapical radiograph of the incisors revealed fully calcified pulp chambers. To prevent iatrogenic perforation of the root canal, the hole was visually and tactiley inspected with a microscope and an ISO 10 Kerr-File plus apex locator (Root ZX, Morita; Kyoto, Japan). The pins were inserted, silica coated (Cojet Sand, 3M Oral Care), silanized (Monobond Plus, Ivoclar Vivadent; Schaan, Liechtenstein), and covered with opaque composite (IPS Empress Direct Opaque, Ivoclar Vivadent). The dental hard tissue was conditioned with an etch-and-rinse bonding agent (Optibond FL, Kerr; Orange, CA, USA) and buildups were made with dentin shades of nanofilled composite (Filtek Supreme XTE A2D, 3M Oral Care). Then, a chamfer preparation was performed (Fig 18).

Figure 19 shows the preparations of the mandibular incisors and premolars, where only the undercut areas have been removed. Additionally, most of the natural interproximal contacts were preserved.

Both jaws were cast with individualized impression trays and polyvinylsiloxane materials (Aquasil Ultra, Dentsply; Milford, CT, USA). A fine impression of the mandibulo-maxillary relationship was made using the above-mentioned composite splint and provisional cement (Temp Bond, Kerr). In the maxillary incisor region, both the splint (Fig 20) and the maxillary overdenture (Fig 21) were cut back to fit the height of the composite buildups. Provisional crowns were made for these teeth with self-curing provisional composite (Protemp Garant, 3M Oral Care) and inserted with provisional cement (Rely X Temp, 3M Oral Care). No other teeth received provisional crowns; they were merely covered with the overextensions, which were then readjusted by relining them with self-curing composite (Protemp Garant, 3M Oral Care) and fixed using of adhesive paste.

The ICRs were made of light-curing composite (SR Adoro, Ivoclar Vivadent) which was finally hardened in an oven at 104°C (Fig 22). When applicable, the restorations were additionally reinforced with a fiber mesh (StickNET, Stick Tech; Turku, Finland). Opaque shades of the material were used in the thin areas of each restoration to mask the dark amber color of the natural teeth. A group function in dynamic occlusion was adjusted by the dental technician to distribute the shear forces that occur during mastication for each restoration.

The 20 restorations were inserted individually on two consecutive days (ten per day). First, the teeth were isolated with gingival retraction cords. 37% phosphoric acid gel was used for conditioning the enamel (30 s) and dentin (15 s), while the composite buildups and the restorations were conditioned by silica coating and silanization. For restoration insertion, dual-curing composite cement (Multilink, Ivoclar Vivadent) was used, and excess material was removed under a microscope using scalers and scalpels. At that point, the overdentures were left off. Occlusion and group function were adjusted by grinding, after which the restorations were polished. Subsequently, the molars were restored in the same manner. Because of the severe destruction of the first molars, parapulpal pins and composite buildups were created before the chamfer preparation was performed. The second molars were prepared minimally invasively (Fig 23).

Oral hygiene was continuously improved at monthly visits with professional tooth cleaning. Three months after the restorations were placed, sound gingival conditions were observed. At the 1.5-year follow-up, bite-wing radiographs were taken, revealing secondary caries in both mandibular second molars due to inadequate hygiene in these interproximal spaces. Both restorations were repaired with composite according to the recommendations of Loomans and Özcan.32 Figures 24 to 27 show the clinical findings 2 years after restoration placement.

**DISCUSSION**

**Pretreatment**

The present case describes the full-mouth rehabilitation with ICRs of an adolescent patient with dentinogenesis imperfecta type II. Typically, these patients present a severe loss of dental hard tissue, as enamel chips off when the teeth achieve occlusion. This can lead to dysgnathia during periods of somatic growth.

In this particular case, the patient had an Angle Class II malocclusion with an increased overjet. Therefore, the patient started orthodontic treatment at the prepubertal stage with functional appliances to stimulate mandibular growth. Because of poor patient compliance, the orthodontic treatment was cancelled after a short duration.

When the patient was 13 years old, she restarted interdisciplinary treatment at our clinic. The first step was increasing vertical occlusion with the overdentures for the first time. Six months after the first bite increase, we needed to raise vertical dimension further to improve the patient’s facial proportions. To obtain maximal intercuspitation in a Class I occlusion with physiological overjet and
overbite, we additionally protruded the mandible forward with an incorporated bite plane. One year after all the restorations had been inserted, however, the bite relapsed to an Angle Class II malocclusion, but the intercuspidation was still sufficient (Figs 25 to 27). Dysgnathia surgery was considered but rejected by the patient and her mother, since the patient was satisfied with her situation.

**Treatment**

In the present case, treatment was performed with indirect composite restorations created using an individual lamination technique. These restorations can be manufactured with a thickness of 0.2 mm; a retentive preparation is not needed, as the restorations are bonded. This allows more healthy hard tissue to be preserved than when conventionally cemented restorations are used. However, thin material layers can reduce the masking ability of the restorations, which can be a concern in some cases. In our patient, some restorations had cervical margins with a thickness of 0.2 mm as a result of the compromise between saving hard tissue and the masking effect. Different thicknesses were previously tested using the diagnostic wax-up, and thicker restoration margins would either have lead to more bulbous crowns or greater loss of hard tissue. As it must be assumed that this young patient would require further full-mouth rehabilitation treatment during her lifetime, saving as much enamel as possible had first priority.

Initially, we considered bleaching the teeth, as there is one case report in the literature that describes successful bleaching in a patient with DI. However, we made no attempt to bleach our patient’s teeth, since it had not been successful in her brother, in whom we had attempted it shortly before.

The histological structure of the enamel in patients with DI has proven to be regular, so a reliable bond between composite and enamel can be ensured. In contrast, the histological structure of the dentin shows less mineralization, more collagen, and lower hardness. Therefore, reduced bond strength between composite and dentin is likely. As some of our patient’s teeth had little or no residual enamel, we did not want to rely on adhesion alone. Therefore, we performed a conventional chamfer preparation for additional mechanical retention in these teeth. To assure the best possible bond strength among dental hard tissue and composite, an etch-and-rinse adhesive system was used. Furthermore, the ICRs and the direct composite buildups were silica coated and silanized, respectively, which is intended to deliver the best adhesion. However,
there is evidence that adhesion to hard tissues affected by DI is compromised due to an increased water content and a considerably reduced mineral content of the dentin. Another study found that it was not possible to obtain a sufficient hybrid layer at the composite-dentin interface. Therefore, we used parapulpal pins to increase retention of the direct buildups. The present authors are aware that parapulpal pins do not deliver the same stability as conventional prosthetic posts, but we wanted to leave this option open for future rehabilitation, if necessary. Finally, the risk of root perforation is high, due to the obliteration of the coronal parts of the root canals (Fig 8). Thus, Pettiette et al suggest performing early prophylactic endodontic treatment before severe root canal obliteration occurs. However, new advances in endodontontology, eg, as guided endodontics, might solve this problem and obviate prophylactic endodontic treatment at this age.

The weak adhesion of enamel to dentin in these patients is the reason for the severe attrition and loss of the vertical dimension. Consequently, Henke et al recommended removing all of the residual enamel to assure crown retention. The authors of the present article are convinced of enamel stabilization when it is fully covered with an adhesive restoration; this is supported by the literature. At present, long-term data on the performance of adhesively bonded restorations in patients with dental malformations, especially dentinogenesis imperfecta, is lacking. Only one study has examined the longevity of composite restorations in patients with molar-incisor hypomineralization, while other authors compared the longevity of both composite and ceramic restorations in patients with amelogenesis imperfecta vs a healthy control group. However, these studies did not describe whether the composite restorations were directly or indirectly made, and it remains unclear whether there might be a difference between these approaches, as shown by Manhart et al. Two studies have shown that restorations in such patients were usually replaced when gingival retraction occurred over time, and the patients wanted restorations to be replaced for esthetic reasons.

Nevertheless, for children or adolescents, most therapies cannot be considered as final and definitive, especially in such challenging cases. However, more invasive techniques can be postponed for as long as possible, according to the dynamic treatment concept. Postoperative elongation, further growth of the jaws, and gingival retraction will make additional rehabilitation treatments necessary, but these will be much easier if the natural teeth have been preserved.

CONCLUSION

In conclusion, ICRs are a valuable tool in the treatment of children and adolescents, as they allow treatment which conserves hard tissue even in challenging cases which require full-mouth rehabilitation.

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Clinical relevance: Laboratory-made composite restorations enable conservation of hard tissue while restoring large defects, because these restorations can be manufactured with very thin lamination strength. Therefore, they should receive increased attention for the treatment of children and adolescents.