Submitted: 06 May, 2023 Accepted: 08 June, 2023 Published: 03 March, 2024 DOI:10.22514/jocpd.2024.047

CASE REPORT

Journal of Clinical Pediatric Dentistry

# Dentinogenesis imperfecta: case report with nanoceramic resin crowns restorative treatment

Maria Dolores Casaña-Ruiz[1], Neus Frechina[2], Filo Estrela[2], Montserrat Catalá-Pizarro[2,*]

[1]Faculty of Medicine and Dentistry, University of Valencia, 46010 Valencia, Spain
[2]Department of Stomatology, Faculty of Medicine and Dentistry, University of Valencia, 46010 Valencia, Spain

***Correspondence**
montserrat.catala@uv.es
(Montserrat Catalá-Pizarro)

**Abstract**

Children with dentinogenesis imperfecta require restorative or prosthodontic treatment to minimize the aesthetic and functional impact of the condition. This clinical case report describes the oral rehabilitation procedure in a 12-year-old patient with dentinogenesis imperfecta type II using nanoceramic resin crowns fabricated with Computer-Aided Design/Computer-Aided Manufacturing (CAD/CAM) technology and the patient's progression over eight years. This minimal intervention approach enabled functional and aesthetic reestablishment along with tooth wear prevention. The result simplified an extensive prosthetic procedure and facilitated an affordable rehabilitation for the young patient while providing excellent long-term outcomes.

**Keywords**

Pediatric dentistry; Dentinogenesis imperfecta; CAD/CAM; Case management

## 1. Introduction

Dentinogenesis Imperfecta (DI) is a dentine disorder that occurs during the histodifferentiation stage of tooth development [1]. It results from an alteration in chromosomal alleles during dentin formation. Moreover, it can occur in isolation as a familial trait and as a component of the skeletal dysplasia Osteogenesis Imperfecta (OI) [2].

Teeth affected by this pathology present varying colors ranging from yellowish brown to bluish brown. Radiographically, bulbous crowns with a noticeable constriction at the cement-enamel junction level, short and thin roots, and partial or total obliteration of the pulp chamber and root canals are typical [3].

Enamel is not affected, but due to the alteration of the cement-enamel junction, it breaks off easily from the incisal edge and the occlusal surface, resulting in marked attrition and consequent loss of vertical dimension [4]. The severity of lesions varies depending on the individual, with different forms of presentation in the same family [5].

Dentinogenesis Imperfecta type II, also called opalescent dentin, presents similarities to Dentinogenesis Imperfecta type I, except that it is not linked to a systemic condition apart from the dental disorder [5]. It is characterized by severe dental anomalies in both temporary and permanent dentitions, as well as an important hereditary association [6].

Prosthetic rehabilitation is usually the first choice of treatment when there is a significant loss of enamel [7]. However, extensive rehabilitation is challenging for young patients as it includes multiple visits and procedures that can exceed children's tolerance and cooperation, thereby compromising the quality and acceptance of the final results [8].

The aim of this paper is to show the evolution after eight years of a patient with dentinogenesis imperfecta treated with nanoceramic resin restorations using CAD/CAM technology.

## 2. Case report

A 12-year-old male patient visited the clinic in 2012 seeking a solution to his functional and aesthetic problems. He had no associated pathologies, allergies or medications. His mother reported that his father, younger brother, grandmother and one paternal aunt had the same dental condition.

The patient showed young permanent dentition, with the second permanent molars and the second right upper premolar yet to erupt. Ricketts cephalometric findings showed a Class II Division I skeleton with large ANB angle difference, retrusive chin, severely proinclined maxillary incisor and anterior open bite. Oral hygiene was deficient, presenting a mild-moderate gingivitis.

Visual inspection revealed an amber-opalescent dental structure with marked attrition throughout all dentition, resulting in a decrease in vertical dimension (Fig. 1).

The patient did not report pain, sensitivity or any signs of dental infection. The orthopantomography and bitewings showed typical characteristics of Dentinogenesis Imperfecta: bulbous crowns, obliterated root canals and cervical constriction (Fig. 2). Due to his history, clinical and radiographic characteristics, the patient was diagnosed with DI type II.

Initially, two possible treatment options were considered. The first option was a combination of preformed metal crowns and ceramic crowns for posterior teeth, and composite restorations for anterior teeth. The second option was minimal intervention rehabilitation using an intra-oral scanner and individual aesthetical crowns with resin nanoceramic-based


  

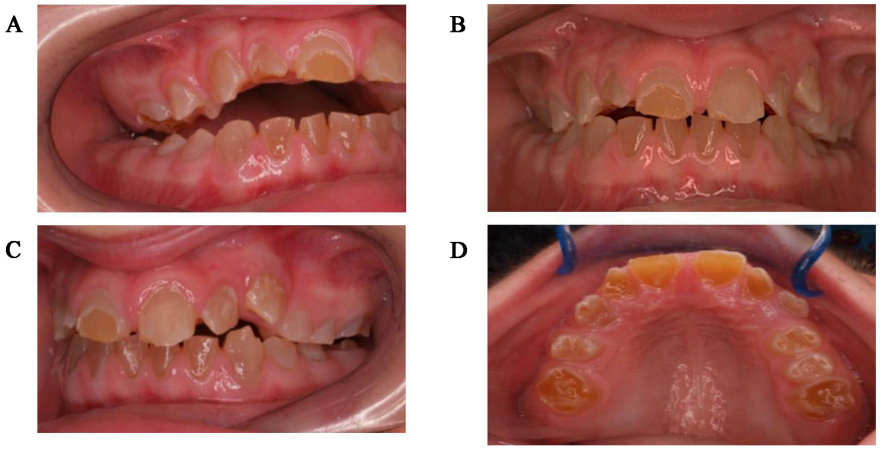


**FIGURE 1. Intra-oral photos.** (A) right lateral view; (B) frontal view; (C) left lateral view; (D) upper oclusal view.


**FIGURE 2. Radiography.** (A) bitewings; (B) orthopantomography.

CAD/CAM material that could be intraorally individualized, repaired and modified if needed. It was proposed that nanoceramic resin crowns would not interfere with dental development neither with planned twin-block treatment.

Finally, the decision was made to restore the vertical dimension, preserving all remaining dental tissue, and recover proper function and aesthetics for the growing child using CAD/CAM technology and the dental chair approach.

In the first stage, alginate impressions (Tropicalgin Chromatic, Zhermack S.p.A., Badia Polesine (RO), Italy) of both upper and lower jaws were taken, and centric relation as well as facial arch were registered. Finally, they were mounted in a semi-adjustable articulator (A7 Plus E, Bio-Art B Soluções Inteligentes, São Carlos, Brazil).

A diagnostic wax-up was conducted to pre-establish the adequate vertical dimension and appropriate dental morphology. In this wax-up, it was considered that the vertical dimension could be increased by 3 mm by adjusting the minimum thickness of the crowns (1.5 mm per unit).

In the second phase, the wax-up was directly scanned through the CEREC® system primescan (Dentsply Sirona; York, Pennsylvania) for the preparation of individual nanoceramic resin crowns (Lava-Ultimate 3M®; Madrid, Spain). A total of twenty-three crowns were produced, four of which corresponded to the first upper and lower molars, three upper premolars and lower ones, four canines, four upper incisors and four lower ones (Fig. 3).

From here on, clinical sessions of adjusting and cementation were planned, starting with posterior teeth, and leaving a period of one month between appointments to allow the child to adapt to the new vertical dimension (Fig. 4). An occlusal reduction was not conducted due to the existing teeth wear. Only angles were rounded, and a path of insertion was slightly marked on buccal surfaces.

In the appointments, once the adjustment of the crown was checked, the internal surface was treated by sanding (CoJet$^{TM}$; 3M ESPE, Madrid, Spain), cleaned with alcohol, and after drying, a universal adhesive with silane was applied, applied by rubbing for 20 seconds, and dried again with air to evaporate the solvent.

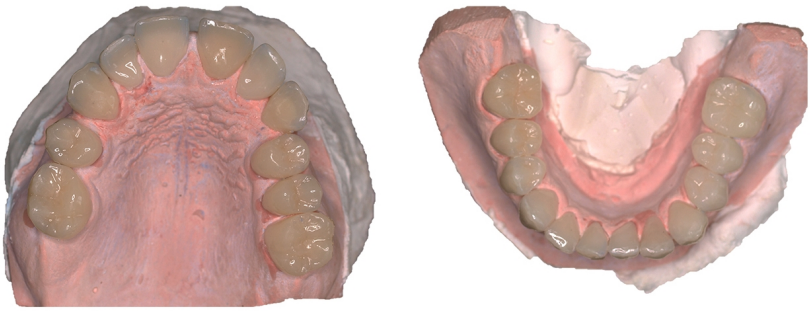

**FIGURE 3. Models with nanoceramic resin crowns.**

**FIGURE 4. Intra-oral photo, posterior crowns cementation.**

Dental structure was etched with orthophosphoric acid for 15 seconds, rinsed with water and slightly dried with air. A layer of photopolymerizable universal adhesive was applied, rubbed for 20 seconds, dried for 5 seconds to evaporate the solvent, and then polymerized for 10 seconds.

As a cement, composite dual (Grandio® Core Dual Cure; Cuxhaven; Germany) was used, polymerizing from different angles for an adequate setting. Finally, marginal adjustment was confirmed with bitewings (Fig. 5), margins were polished, and occlusion was checked.

## 3. Results

When the patient was 13 years old, check-up appointments took place every three months. At the same time, orthopedic treatment with twin-block appliance was started to improve the jaw position (mandibular retrognathia) and dentofacial discrepancy. After one year, it was observed that the crown integrity was maintained, and no functional or aesthetic problems had occurred. The child and the parents were all very satisfied with the restorative treatment.

In successive visits, the importance of correct oral hygiene was highlighted, both to the patient and to his parents, and the need for flossing was explained. From the first year onwards, follow-up check-ups were scheduled every six months.

In December 2014 (14 years old), after reevaluation by the orthodontists, two teeth were extracted to create space for alignment and improve the overjet, the first being the upper right first molar to allow eruption of the blocked second upper right premolar, and the second being the upper left second premolar. The lower right first premolar crown was repaired once due to a fracture and finally had to be replaced after three years. Moreover, a new crown was made for the upper right second premolar.

The second stage of orthodontic treatment began in 2015, when the patient was 15 years old, with the purpose of correcting the overbite and thereby enable dental alignment. In January 2018 (18 years old), it was necessary to extract the lower left first premolar due to significant caries destruction and the impossibility of restoring the tooth. Rehabilitation of second permanent molars was postponed due its integrity and impaction of mandibular third molars.

During a routine appointment in September 2020 (20 years old), clinical exploration revealed supragingival dental calculus in the lower incisor area, with mild gingivitis. Radiographically, caries was diagnosed in the lower left and right first molars and a loose crown in the upper right second premolar and lower left second premolar. In addition, the lower third molars were suspected of being impacted. The rest of the restorations maintained adequate function and aesthetics with a stable vertical dimension.

At this point (Figs. 6,7), the 20-year-old patient was informed about the possibility of planning a rehabilitation and replacing resin crowns with more long-lasting and aesthetic materials. The patient was referred to the University's Prosthetics department, where he underwent a complete dental restoration using ceramic materials.

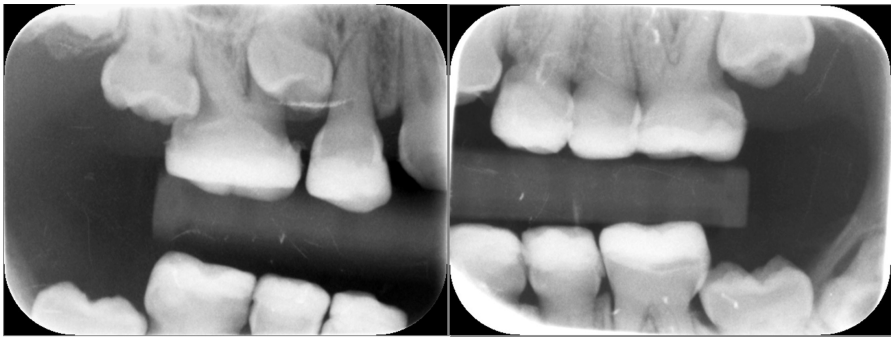

**FIGURE 5. Bitewings with cemented crowns.**

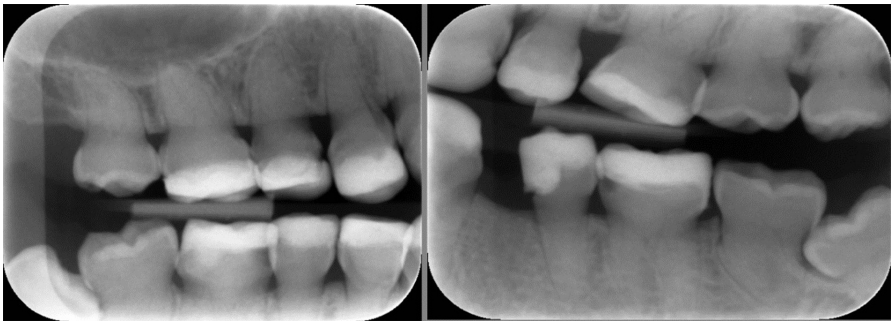

**FIGURE 6. Bitewings at eight years follow-up.**

**FIGURE 7. Intra-oral photos, patient's evolution eight years follow-up.**

## 4. Discussion

Dentinogenesis imperfecta type II (DI-II) has an estimated prevalence of 1:6000 to 1:8000 regardless of sex or race [9]. Histologically, it consists on disorganized dentine with irregularly set dentinal tubules and a decalcified matrix surface. Tubules tend to be wider in diameter and occur in lower numbers in comparison with normal dentine [10].

Enamel, although normal in appearance, usually presents microfractures or fissures and tends to break off. This is due to a weak cement-enamel junction, exposing abnormally soft dentin, which causes rapid dental wear [11].

As tooth wear and discoloration progress, there is an even greater need to treat and restore aesthetics and function early. This aesthetic concern, observed in most cases, has a negative impact on the patient's self-esteem, affecting their interpersonal relationships. Moreover, it has been shown to be directly related to their quality of life, regardless of the age group [12].

A wide variety of treatment options exist, all oriented towards preventing fast enamel and dentine wear and improving tooth aesthetics [13]. Depending on the severity of the case, possible alternatives include conventional composites, aesthetic crowns in anterior teeth, preformed crowns in posterior teeth, metal-ceramic crowns, resin crowns, overdentures or removable devices. Treatment selection will depend on other factors, such as adhesion problems, lack of cooperation in pediatric patients, the complexity of restoring procedures and fracture tendency. The low fracture resistance of DI-II affected teeth can be attributed to the poorly woven microstructure of their dentin, which leads to a reduction in hardness [13].

Regarding adhesive techniques, most adhesives now achieve stable dentinal junctions compared to the "gold standard" particularly when working with self-etching. It has been observed that applying universal adhesives on dentine should not be preceded by phosphoric acid etching since there is no improvement in the restoration loss average [14].

Thus, applying self-etching seems to be the best option in these cases for dentinal adhesion, completing procedures with reduced times [15].

Considering histological particularities of DI patient's dentin, it is possible to use additional treatments to improve the surface condition and achieve good adhesion. For example, the sanding technique (air abrasion) used for ceramics allows preparing the crown's surface, creating superficial micro-roughness that enhances retention energy [16].

On the other hand, using cements with adhesive potential, as well as applying biomaterials containing 10-MDP or 10-Methacryloyloxydecyl dihydrogen phosphate, are two other alternatives to consider. Particularly, the latter is one of the few monomers used in dental adhesion that has shown a chemical binding to dental tissue through ionic bonds with calcium found in hydroxyapatite [16].

Classically, the recommendation when treating patients affected by DI, was to remove all residual enamel to ensure crown retention [17].

Current practices aim to preserve enamel, covering affected teeth with adhesive restorations [18].

The use of nanoceramic resin crowns would be a good option for young permanent teeth not only for DI, but also for extensive caries or decalcifications, enamel defects or difficulty in controlling humidity. Preservation of the remaining tooth structure is essential for the complete root development, as well as for the establishment of the occlusion, to consider together with direct and indirect composites. But in the case of patients with DI, requirements vary and the degree of adhesion is altered, so each treatment option should be carefully considered [19].

Creating crowns using CAD/CAM offers significant advantages over conventional techniques, including faster clinical practice, an improved experience for the patient, and the quality of the restorations. On the other hand, the main limitations are the cost of material, the cost of equipment and the additional costs that may be incurred [20].

When the preparation and clinical success depend on mechanical friction, digital impressions are an appropriate option. In this case, however, the objective was to prepare the insertion pathways, and this was successfully achieved with the alginate impressions [20].

The minimal intervention technique used for the restorations did not require the classical tooth preparation with any type of gingival finishing line. The material used, based on nanoceramic resins, has allowed to carry out the oral rehabilitation in cases of young permanent dentition with minimal dental preparation. This type of resin is composed of 80% nanoceramic particles, which confer high fracture and flexion resistance, great resilience (no wear), and aesthetic properties (translucency and fluorescence) similar to natural teeth. Additionally, all preparation and design can be conducted at the clinic, which saves time between appointments and implies a reduction in laboratory costs. Likewise, given that no baking is needed, they can be repaired or modified *in situ* if needed. As alternative materials for treatment, hybrid resin blocks where resin composite is combined with ceramic particles can be considered. They are characterized by their precision, strength, flexibility and guarantee a long-lasting brightness after restoration [20].

Therefore, adhesive reconstruction preserves tooth structure and is a predictable approach for good esthetics, posterior occlusion and patient's adaptation to the new VD (Vertical Dimension) [21].

One of the strengths of this paper is the long-term follow up of 8 years, but even though the results in our patient are successful, most therapies cannot be considered definitive for children and teenagers, especially in such challenging cases. Post-eruptive lengthening, a bigger maxillary growth, and passive eruption make additional rehabilitation treatments necessary. These will be more favorable if the remnant dental structure has been conserved [21].

If we compare our findings with those of other authors, most of the latter preserve the remaining tooth structure. The placement of metal crowns in posteriors and anterior composite restorations are the most widespread practice. However, cases of dental extractions and placement of partial dentures have also been found [22].

In terms of follow-up time, regular check-ups are essential. Treatments are usually conducted at two points in time: mixed dentition and permanent dentition. Patients with DI are usually

followed up until adulthood and the cessation of growth [22].

In adulthood, patients affected by DI usually require multidisciplinary treatment. In moderate-severe cases, prosthetic restorations dominate compared to mild cases, where composite restorations are more common [23].

The use of technology (CAD-CAM) allows the creation of high-precision dental devices and prostheses that simplify the planning and execution of advanced implant surgery and full-mouth rehabilitation.

Due to higher aesthetic requirements, the use of tools such as DSD (Digital Smile Design) is also highly recommended. The advantage of preserving the remaining tooth structure is to delay or even avoid the need for mucosa- or implant-supported prostheses.

## 5. Conclusions

Using nanoceramic resins with CAD/CAM technology has enabled non-invasive oral rehabilitation, providing significant advantages at an early age. Therefore, it has been possible to restore the dental structure, offering functional treatment, improving the patient's aesthetics and quality of life.

Despite the prejudices of this adhesion-based technique could raise in dentinogenesis imperfecta, the results in our case are hopeful and supports its use.

Eight years after treatment, the patient still has the crowns, indicating a long-term provisional solution.

### ABBREVIATIONS

DI, dentinogenesis imperfecta; DI-II, dentinogenesis imperfecta type II; OI, osteogenesis imperfecta; CAD/CAM, Computer-Aided Design/Computer-Aided Manufacturing; VDO, vertical dimension.

### AVAILABILITY OF DATA AND MATERIALS

The data presented in this study are available on reasonable request from the corresponding author.

### AUTHOR CONTRIBUTIONS

MCP and FE—designed the research study. NF and MDCR—performed the research. MDCR—provided help and advice. All authors contributed to editorial changes in the manuscript. All authors read and approved the final manuscript.

### ETHICS APPROVAL AND CONSENT TO PARTICIPATE

Due to local regulations of Lluis Alcanyís Foundation. Dental Clinic of the University of Valencia (Health Center 5452), no ethical approval is required for this retrospective case report. An informed consent, for treatment and scientific diffusion of the case was signed by the parents and the patient, respectively.

### ACKNOWLEDGMENT

Not applicable.

### FUNDING

This research received no external funding.

### CONFLICT OF INTEREST

The authors declare no conflict of interest.

### REFERENCES

[1] Mckinney R, Olmo H. Developmental Disturbances of the teeth, anomalies of structure. In StatPearls. StatPearls Publishing: Treasure Island (FL). 2022.

[2] Stephen L, Beighton P. Dental management of severe dentinogenesis imperfecta in a mild form of osteogenesis imperfecta. Journal of Clinical Pediatric Dentistry. 2003; 26: 131–136.

[3] Neri Morales C, Silva Amaro A, Cardona JD, Bendeck JL, Cifuentes Gaitan K, Ferrer Valencia V, *et al*. Osteogenesis imperfecta: a case series and literature review. Cureus. 2023; 8: e33864.

[4] Santos-Hidalgo-Gato G, Enriquez-Hidalgo Y, Coto-Valladares LM. Comprehensive diagnosis and treatment of amelogenesis imperfecta. MediSur. 2021; 20: 7–9.

[5] Grover C, Dhawan P, Kaur H, Kakar A. Osteogenesis imperfecta and dentinogenesis imperfecta: clinical features and dental management. Current Medicine Research and Practice. 2022; 12: 167–172.

[6] Soliman S, Meyer-Marcotty P, Hahn B, Halbleib K, Krastl G. Treatment of an adolescent patient with dentinogenesis imperfecta using indirect composite restorations—a case report and literature review. The Journal of Adhesive Dentistry. 2018; 20: 345–354.

[7] Azpiazu-Flores FX, Knobloch LA, Larsen PE. Interdisciplinary management of a patient with dentinogenesis imperfecta type II using a combination of CAD-CAM and analog techniques: a clinical report. Journal of Prosthodontics. 2022; 31: 647–654.

[8] Millet C, Ducret M, Khoury C, Virard F. Monolithic CAD/CAM complete overdentures for a pedodontic patient with dentinogenesis imperfecta and limited prosthetic space: a clinical report. The International Journal of Prosthodontics. 2020; 33: 341–346.

[9] Desai V, Chitguppi R. Nonsyndromic dentin genetic diseases: dentinogenesis imperfecta type III: a unique presentation of rhizomegaly, taurodontism, and dilacerated roots. Saudi Journal of Oral Sciences. 2021; 8: 184–188.

[10] Gallusi G, Libonati A, Campanella V. SEM-morphology in dentinogenesis imperfecta type II: microscopic anatomy and efficacy of a dentine bonding system. European Journal of Paediatric Dentistry. 2006; 7: 9–17.

[11] Du Q, Cao L, Liu Y, Pang C, Wu S, Zheng L, *et al*. Phenotype and molecular characterizations of a family with dentinogenesis imperfecta shields type II with a novel *DSPP* mutation. Annals of Translational Medicine. 2021; 9: 1672.

[12] Garrocho-Rangel A, Dávila-Zapata I, Martínez-Rider R, Ruiz-Rodríguez S, Pozos-Guillén A. *Dentinogenesis imperfecta* type II in children: a scoping review. Journal of Clinical Pediatric Dentistry. 2019; 43: 147–154.

[13] Beltrame APC, Rosa MM, Noschang RA, Almeida IC. Early rehabilitation of incisors with dentinogenesis imperfecta type II—case report. Journal of Clinical Pediatric Dentistry. 2017; 41: 112–115.

[14] Appelstrand SB, Robertson A, Sabel N. Patient-reported outcome measures in individuals with amelogenesis imperfecta: a systematic review. European Archives of Paediatric Dentistry. 2022; 23: 885–895.

[15] Maciel Pires P, Dávila-Sánchez A, Faus-Matoses V, Nuñez Martí JM, Lo Muzio L, Sauro S. Bonding performance and ultramorphology of the resin-dentine interface of contemporary universal adhesives. Clinical Oral Investigations. 2022; 26: 4391–4405.

[16] Massé L, Etienne O, Noirrit-Esclassan E, Bailleul-Forestier I, Garot E. Dentine disorders and adhesive treatments: a systematic review. Journal of Dentistry. 2021; 109: 103654.
[17] Guichet DL. Digital workflows in the management of the esthetically discriminating patient. Dental Clinics of North America. 2019; 63: 331–344.
[18] Velásquez Baena L, Restrepo Restrepo M, Mejía Roldan JD. Indirect restorations for the management of hypomineralization of molars and incisors: a conservative approach. Revista De Odontopediatría Latinoamericana. 2023; 13: 423584.
[19] Rolim TZC, da Costa TRF, Wambier LM, Chibinski AC, Wambier DS, da Silva Assunção LR, *et al*. Adhesive restoration of molars affected by molar incisor hypomineralization: a randomized clinical trial. Clinical Oral Investigations. 2021; 25: 1513–1524.
[20] Sarapultseva M, Leleko A, Sarapultsev A. Case report: rehabilitation of a child with dentinogenesis imperfecta with CAD/CAM approach: three-year follow-up. Special Care in Dentistry. 2020; 40: 511–518.
[21] Bouvier D, Leheis B, Duprez JP, Bittar E, Coudert JL. Dentinogenesis imperfecta: long-term rehabilitation in a child. Journal of Dentistry for Children. 2008; 75; 192–196.
[22] Alrashdi M, Schoener J, Contreras CI, Chen S. Full mouth rehabilitation of two siblings with dentinogenesis imperfecta type II using different treatment modalities. International Journal of Environmental Research and Public Health. 2020; 17: 7029.
[23] Thuesen KJ, Gjørup H, Hald JD, Schmidt M, Harsløf T, Langdahl B, *et al*. The dental perspective on osteogenesis imperfecta in a Danish adult population. BMC Oral Health. 2018; 18: 175.

**How to cite this article:** Maria Dolores Casaña-Ruiz, Neus Frechina, Filo Estrela, Montserrat Catalá-Pizarro. Dentinogenesis imperfecta: case report with nanoceramic resin crowns restorative treatment. Journal of Clinical Pediatric Dentistry. 2024; 48(2): 189-195. doi: 10.22514/jocpd.2024.047.