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CASE REPORT:

Association of Osteogenesis Imperfecta with Pre-eruptive Resorption: Two Cases

Osteogénesis imperfecta y reabsorción preeruptiva: dos casos

Melek Rutbil DDS, Research Assistant[1] https://orcid.org/0009-0005-6461-1729
Deniz Ozkara DDS, Research Assistant[1] https://orcid.org/0009-0002-3937-8521
Fatma Akkoca DDS, Assistant Professor[1] https://orcid.org/0000-0002-4522-656X
Melih Ozdede DDS, Associate Professor[1] https://orcid.org/0000-0002-8783-802X

[1]Department of Dentomaxillofacial Radiology, Faculty of Dentistry, Dokuz Eylul University, Izmir, Turkey.

Correspondence to: Melek Rutbil - rutbil.melek@deu.edu.tr

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ABSTRACT: Osteogenesis imperfecta (OI) is a rare hereditary disorder of the connective tissue, characterized by bone fragility and associated craniofacial and dental abnormalities. Pre-eruptive intracoronal resorption (PIR) is an uncommon developmental anomaly that affects unerupted teeth, typically presenting as asymptomatic radiolucent lesions within the coronal dentin. Although both conditions have been individually described in the literature, their coexistence has not been previously reported. This case report describes two male patients, aged 45 and 51 years, both diagnosed with OI and found to have PIR in multiple impacted teeth. In the first case, panoramic radiographs revealed radiolucent lesions within the crowns of impacted teeth #13 and #33, which were further characterized by cone-beam computed tomography (CBCT) as PIR involving enamel and dentin. In the second case, panoramic imaging demonstrated PIR in impacted teeth #37, #38, and #48, confirmed by CBCT as extensive intracoronal defects adjacent to the mandibular canal. Both patients were referred for surgical evaluation following radiographic diagnosis. This report highlights the potential coexistence of OI and PIR, raising questions about possible shared pathogenic mechanisms. More importantly, the cases emphasize the role of radiographic evaluation in early diagnosis.

KEYWORDS: Collagen diseases; Cone-beam computed tomography; Osteogenesis imperfecta; Panoramic radiography; Teeth abnormalities; Tooth resorption.

RESUMEN: La osteogénesis imperfecta (OI) es un trastorno hereditario raro del tejido conectivo, caracterizado por la fragilidad ósea y asociado con anomalías craneofaciales y dentales. La reabsorción intracoronal preeruptiva (RIP) es una anomalía del desarrollo poco común que afecta a los dientes no erupcionados, presentándose típicamente como lesiones radiolúcidas asintomáticas dentro de la

dentina coronal. Aunque ambas condiciones han sido descritas individualmente en la literatura, su coexistencia no ha sido previamente reportada. Este informe describe dos pacientes masculinos, de 45 y 51 años de edad, ambos diagnosticados con OI y con presencia de RIP en múltiples dientes impactados. En el primer caso, las radiografías panorámicas revelaron lesiones radiolúcidas en las coronas de los dientes impactados #13 y #33, las cuales fueron caracterizadas mediante tomografía computarizada de haz cónico (CBCT) como RIP que involucraba esmalte y dentina. En el segundo caso, las imágenes panorámicas mostraron RIP en los dientes impactados #37, #38 y #48, confirmadas por CBCT como defectos intracoronales extensos adyacentes al conducto mandibular. Ambos pacientes fueron remitidos para evaluación quirúrgica tras el diagnóstico radiográfico. Este informe resalta la posible coexistencia de OI y RIP, planteando interrogantes sobre mecanismos patogénicos compartidos. Más importante aún, los casos enfatizan el papel de la evaluación radiográfica en el diagnóstico temprano.

PALABRAS CLAVE: Enfermedades del colágeno; Tomografía computarizada de haz cónico; Osteogénesis imperfecta; Radiografía panorámica; Anomalías dentarias; Reabsorción dental.

INTRODUCTION

Osteogenesis imperfecta (OI), commonly known as brittle bone syndrome, is a heritable connective tissue disorder with an estimated prevalence of 1 in 15,000-20,000 live births (1). Most cases result from pathogenic variants in the COL1A1 or COL1A2 genes, which are responsible for type I collagen synthesis (1). Since type I collagen is the principal structural component of bone, skin, tendons, ligaments, and dental tissues, OI manifests not only with skeletal fragility but also with multiple craniofacial and dental complications (2-4).

Pre-eruptive intracoronal resorption (PIR) is a rare developmental anomaly that most frequently affects unerupted mandibular molars (5). Its etiology remains unclear, but proposed contributing factors include trauma, infection, genetic predisposition, and disturbances in tooth development (6). The clinical significance of PIR lies in its typically asymptomatic course until eruption, when progressive resorptive lesions may lead to pulpal involvement and complex treatment needs (5,6).

Although craniofacial and dental abnormalities are frequently observed in patients with OI, no previous reports have described a possible association between OI and PIR (7). This gap highlights the clinical and academic importance of exploring their potential coexistence.

Compared to conventional radiography, cone-beam computed tomography (CBCT) provides superior three-dimensional visualization, offering high diagnostic accuracy for developmental anomalies such as PIR (8). CBCT allows precise localization of resorptive lesions, detailed characterization of their morphology, and accurate assessment of their spatial relationships with adjacent structures such as the mandibular canal and neighboring roots (9,10). These features make CBCT an essential tool for diagnosis, treatment planning, and surgical decision-making in complex maxillofacial cases (8-10).

The aim of this case report is to evaluate the coexistence of OI and PIR and to emphasize the importance of panoramic radiography and CBCT in their diagnosis.

CASE REPORTS

CASE 1

A 45-year-old male patient presented to the Department of Oral Diagnosis and Dentomaxillofacial Radiology for routine dental examination. His medical history was significant for OI. Panoramic and periapical radiographs revealed well-defined, round-to-oval radiolucent lesions within the coronal dentin of impacted teeth #13 and #33, raising suspicion of PIR (Figure 1.A, Figure 1.B, Figure 1.C). Consequently, CBCT was performed for further evaluation. The CBCT images were acquired using a Planmeca ProMax 3D Max (Planmeca Oy, Helsinki, Finland) unit. For Case 1, the scans were obtained at standard dose settings (96 kVp, 5.6 mA, 12.1 s), with a field of view (FOV) of 10 × 9 cm and voxel size of 200 µm. For Case 2, the same device was used with identical exposure parameters (96 kVp, 5.6 mA, 12.1 s), with a FOV of 13 × 5.5 cm and voxel size of 200 µm.

CBCT demonstrated that tooth #13 was impacted in a semi-vertical position, with its crown oriented disto-palatally and its root directed mesio-buccally. The root was in proximity to the nasal floor, while the crown was adjacent to the root of tooth #14. A Type IV PIR involving both enamel and dentin was identified in the crown of tooth #13 (Figure 1.B, Figure 1.D). The diagnostic classification of PIR lesions followed the criteria proposed by Ozden *et al.*, where Type IV PIR refers to lesions extending through both enamel and dentin (11).

Tooth #33 was impacted in a vertical position, in direct contact with the mandibular incisive canal and close to the mental foramen. A Type IV PIR was also detected in its crown. The patient was referred to the Department of Oral and Maxillofacial Surgery for surgical extraction (Figure 1.C, Figure 1.E).

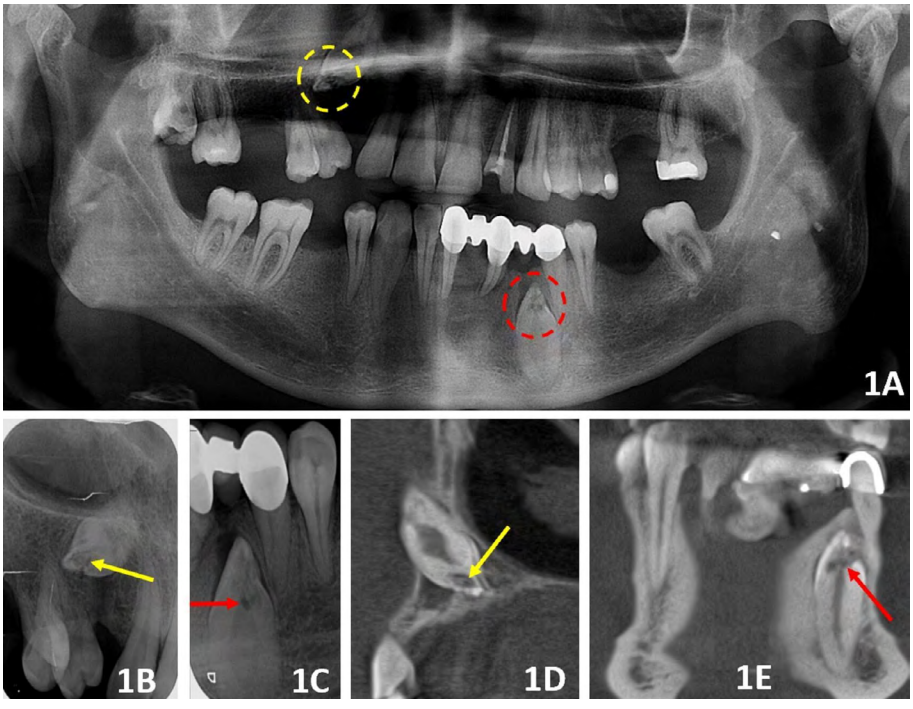

Figure 1. (1A) Panoramic image showing unerupted teeth #13 (yellow) and #33 (red) with pre-eruptive coronal resorption. Periapical view of #13 (1B) and #33 (1C). CBCT sagittal section of #13 (1D) and coronal section of #33 (1E). All images indicate the respective resorption areas with arrows.

CASE 2

A 51-year-old male patient presented to the Department of Oral Diagnosis and Dentomaxillofacial Radiology with a chief complaint of pain in the left mandibular posterior region and a mobile tooth. His medical history was notable for OI, hypertension, chronic obstructive pulmonary disease, and aortic enlargement.

Clinical and radiographic examination revealed that the source of pain was tooth #36, which exhibited mobility, alveolar bone loss in the furcation area, and distal root resorption. The panoramic radiograph also demonstrated radiolucencies involving the crowns of impacted teeth #37, #38, and #48 (Figure 2.A).

CBCT examination was performed to further assess the PIR lesions and their relationship with the mandibular canal before extraction (Figure 2.B, Figure 2.C, Figure 2.D). Tooth #48, a two-rooted molar, was impacted in a mesioangular position. A Type IV PIR lesion affecting both enamel and dentin was observed in the mesial crown of this tooth (Figure 2.B).

Tooth #37, a two-rooted molar, was impacted in a mesioangular position, with the mandibular canal located inferior and lingual to its roots. A Type IV PIR involving both enamel and dentin was identified in its crown (Figure 2.C).

Tooth #38, a single-rooted molar, was also impacted mesioangularly, with the mandibular canal positioned lingual to its root. A PIR lesion affecting the enamel was observed in the mesial crown of this tooth, which was in proximity to impacted tooth #37 (Figure 2.D).

The patient, who was under regular medical follow-up, was referred to the Department of Oral and Maxillofacial Surgery for extraction of the symptomatic tooth #36 and the impacted teeth affected by PIR.

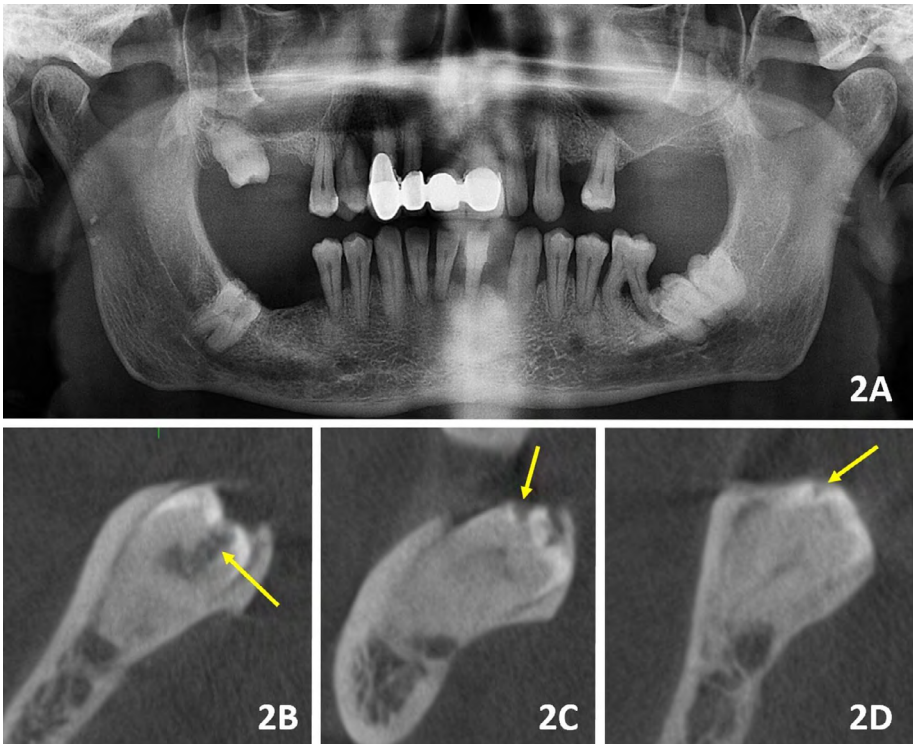

Figure 2. (2A) Panoramic image showing pre-eruptive coronal resorption in unerupted teeth #48, #37, #38, and a lesion in #36. CBCT cross-sectional views of #48 (2B), #37 (2C), and #38 (2D), all demonstrating resorption areas.

DISCUSSION

This case report presents two patients diagnosed with both OI and PIR, a combination not previously documented in the scientific literature. The co-occurrence of these conditions raises questions about a possible genetic correlation. However, further clinical and genetic research is needed to establish any definitive association and examine the underlying molecular mechanisms.

OI is most commonly caused by pathogenic variants in the COL1A1 and COL1A2 genes, which encode the α1 and α2 chains of type I collagen (12). Defects in collagen synthesis or structure can result in either quantitative deficiency or qualitative abnormalities (13). Since type I collagen constitutes approximately 90% of the organic dentin matrix, such mutations predispose dentin to resorptive changes by compromising its structural integrity, cross-linking, and mineral-binding properties (7,13,14). In patients with OI, this defective collagen causes adverse modifications of the dentin matrix, poor mineralization density, and abnormal fibrillar structure (2,3). Circumpulpal dentin is inherently weaker than mantle dentin, resulting in spontaneous tooth fractures and enamel loss (7). This fundamental structural compromise provides a reasonable mechanism for increased PIR susceptibility.

Similar associations have been reported in other hereditary dental disorders (15,16). For example, amelogenesis imperfecta has been linked to higher prevalence of PIR, with cases of intracoronal resorption documented in affected patients (15,16). These observations suggest that genetic defects compromising tooth structure may increase susceptibility to resorptive processes, supporting the hypothesis of a shared pathogenic pathway.

This suggests that mutations in COL1A1/COL1A2 may make dentin matrices more vulnerable to enzymatic degradation during the pre-eruptive phase, thus predisposing patients with OI to PIR. Although this remains uncertain, further molecular and clinical studies are needed to clarify the relationship between OI and PIR. From a clinical perspective, our cases emphasize the importance of comprehensive radiographic assessment in OI patients. CBCT provides superior three-dimensional visualization of resorptive lesions compared with conventional radiography, allowing early detection and more accurate treatment planning (8). Regular radiographic surveillance, particularly in patients with severe OI phenotypes, may help identify PIR before significant crown destruction occurs (8).

Dental practitioners managing adult patients with systemic collagen disorders, particularly OI, should perform routine radiographic monitoring of unerupted and impacted teeth, even in the absence of symptoms. When radiolucent intracoronal lesions are observed on panoramic radiographs, CBCT evaluation is advised to assess the extent of resorption and plan appropriate treatment. Early detection allows intervention before pulpal involvement or crown fracture occurs. Close collaboration among oral radiologists, surgeons, restorative dentists, and the patient's medical team is essential for safe and effective management.

CONCLUSIONS

The coexistence of OI and PIR, reported here for the first time, suggests a shared genetic pathway related to type I collagen defects. This finding highlights need for the regular radiographic monitoring of OI patients, with CBCT offering superior detection of pre-eruptive anomalies. Future molecular studies are required to clarify the prevalence and genetic mechanisms, which may guide clinical management and improve understanding of collagen-related dental pathologies.

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