

Case report



INVASIVE CERVICAL RESORPTION OF UNCOMMON ORIGIN: A CASE REPORT WITH 14-YEAR FOLLOW-UP

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ABSTRACT

Background: Invasive cervical resorption (ICR) is a complex, relatively uncommon, and insidious phenomenon. Its occurrence is associated mainly with local predisposing factors, but systemic risk factors may be involved as well. An ICR lesion that develops in the absence of an identifiable cause is termed ‘*idiopathic*’.

Case report: This study presents the surgical management and very long-term follow-up results of an “*idiopathic*” Class 3 ICR lesion in the maxillary central incisor. The case provides a unique insight into an uncommon ICR lesion that exhibited an unusual appearance and features. For the repair material, a sandwich technique was applied using mineral trioxide aggregate (MTA) covered with glass ionomer cement (GIC). At the 14-year follow-up, the tooth was symptom-free and functional; however, the disadvantages of both materials were clearly demonstrated. From that perspective, the study highlighted the discoloration potential of the white form of MTA-Angelus and the lack of regenerative properties of the GIC used. Remarkably, 12 years after the surgery, the patient was diagnosed with Familial Mediterranean fever, a rare systemic disease. However, the connection between this genetic disease and the initiation of ICR remains an open question for now.

Conclusion: The clinician should be alert for the occurrence of ICR lesions, especially in patients with concomitant systemic disorders. The right choice of appropriate repair material is a key factor for the healing outcome of ICR lesions in the anterior zone.

Keywords: invasive cervical resorption, idiopathic, Mineral trioxide aggregate, long-term follow-up, Familial Mediterranean fever,

BACKGROUND

Invasive cervical resorption (ICR) is a complex, inflammatory, and relatively uncommon form of external root resorption. This aggressive phenomenon develops due to damage to the periodontal ligament and subepithelial cementum and osteoclastic activity. It starts and manifests in the tooth’s cervical area, below the epithelial junction, and above the alveolar ridge crest. The resorptive process is dynamic and mainly involves radicular hard dental and periodontal pulpal tissues. If not promptly treated, it may even lead to tooth loss. Early diagnosis and appropriate management have been shown to improve the likelihood of tooth retention [1-3].

The prevalence rate of this disorder is low and ranges from 0.02% to 2.3% [1, 4-6]. The distribution of ICR is mainly associated with maxillary anterior teeth [1, 4, 7, 8]. While the etiology of ICR is not fully elucidated, its occurrence is associated with two major groups of potential predisposing factors: **local (dental-related) factors** and **systemic risk factors** [4, 9]. The impact of local predisposing factors on the initiation of ICR is being investigated more fundamentally. Despite reported minor differences in the distribution pattern of external cervical resorption by odontogenic causal factors, it can be summarized that orthodontic treatment (15.87-45.7%) and trauma (15.1-33.33%) are the most critical among them [4, 5, 6, 8].

Many systemic syndromes, diseases, and conditions have also been reported to be associated with ECR, such as Gaucher’s disease, hereditary haemorrhagic telangiectasia, Paget’s disease, Goltz syndrome, Papillon–Lefèvre syndrome, Stevens–Johnson syndrome, Turner syndrome, osteogenesis imperfecta, hormonal disturbance and fluctuations, bone dysplasia, diabetes, renal diseases, rheumatoid arthritis, systemic sclerosis, medication-induced resorption (bisphosphonates, denosumab), chemotherapy, exposure to viral infections, etc. [3, 5, 9-12]. The list of suspected diseases is gradually increasing, but the causal relationship between some of them and ECR and their role as initiating or contributing factors remains unclear.

External cervical root resorption that develops without a known cause is termed “*idiopathic*.” The definition of idiopathic ICR has evolved with an improved understanding of potential etiologic factors, leading to an up-

dated list [5, 12]. Nevertheless, the term ‘idiopathic resorption’ is still used for numerous clinical cases [9, 13, 14]. Therefore, this study aims to present a very long-term follow-up of a case of ICR with unknown local odontogenic risk factors and a probable, albeit unproven, systemic risk factor.

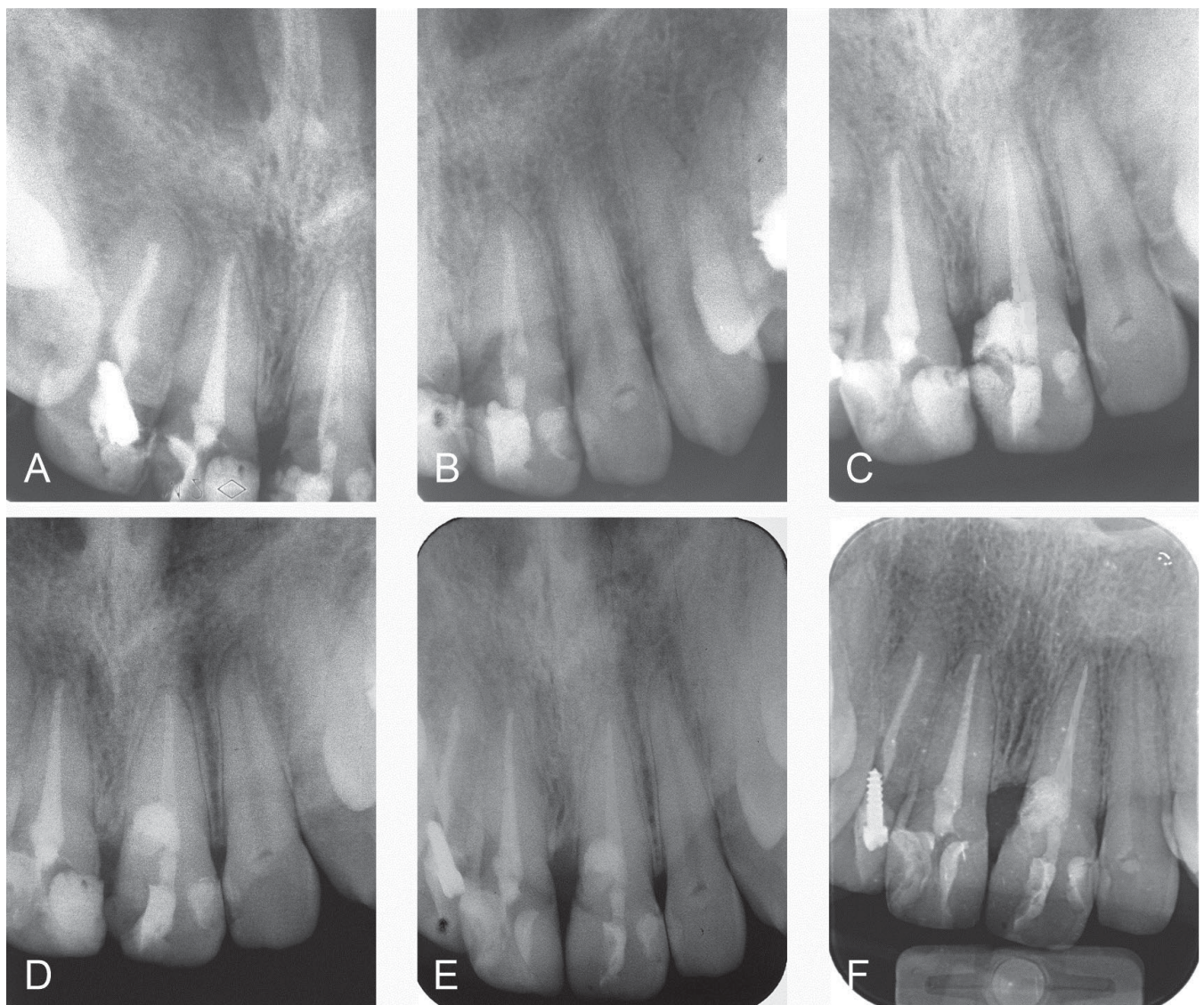
CASE DESCRIPTION

History of the present illness

A 31-year-old female patient, originally from Armenia, presented to the endodontic clinic of the Faculty of Dental Medicine, Sofia, with a chief complaint of unsightly fillings in her upper anterior teeth. Clinical examination revealed large old resin composite restorations with an unaesthetic appearance and rough surfaces. The periodontal tissues in this area showed slight gingival inflammation, especially the papilla between the central incisors (Fig. 2A). The localized gingival hyperplasia in this

area rendered probing painful and problematic, and it also revealed a slight increase in probing depth. Additionally, there was a small displacement to the right of the maxillary midline, and further details about the past dental history were requested in this regard. The patient, however, denied having ever experienced trauma. According to her, this abnormality was congenital, and no orthodontic therapy was performed. Radiographs revealed previous endodontic treatments on some teeth that were clinically asymptomatic. The left central incisor, in particular, showed unsatisfactory root canal filling and a subcrestal radiolucent area with irregular outlines apical to the mesial restoration. The lesion seemed to engage the coronal third of the root and the root canal filling space (Fig. 1A, B), but the defect was not accessible on probing. A deep subgingival carious lesion or an iatrogenic cervical perforation committed during the previous endodontic treatment was initially suspected.

Fig. 1. A) and B) Initial radiographs of tooth 21 taken at different angulations, revealing irregular subcrestal radiolucency apical to the mesial restoration. C) An unsuccessful attempt to manage the lesion internally with MTA. D) Immediate postoperative radiograph of the defect repaired with white MTA-Angelus. E) 3-year follow-up. F) 14-year follow-up.



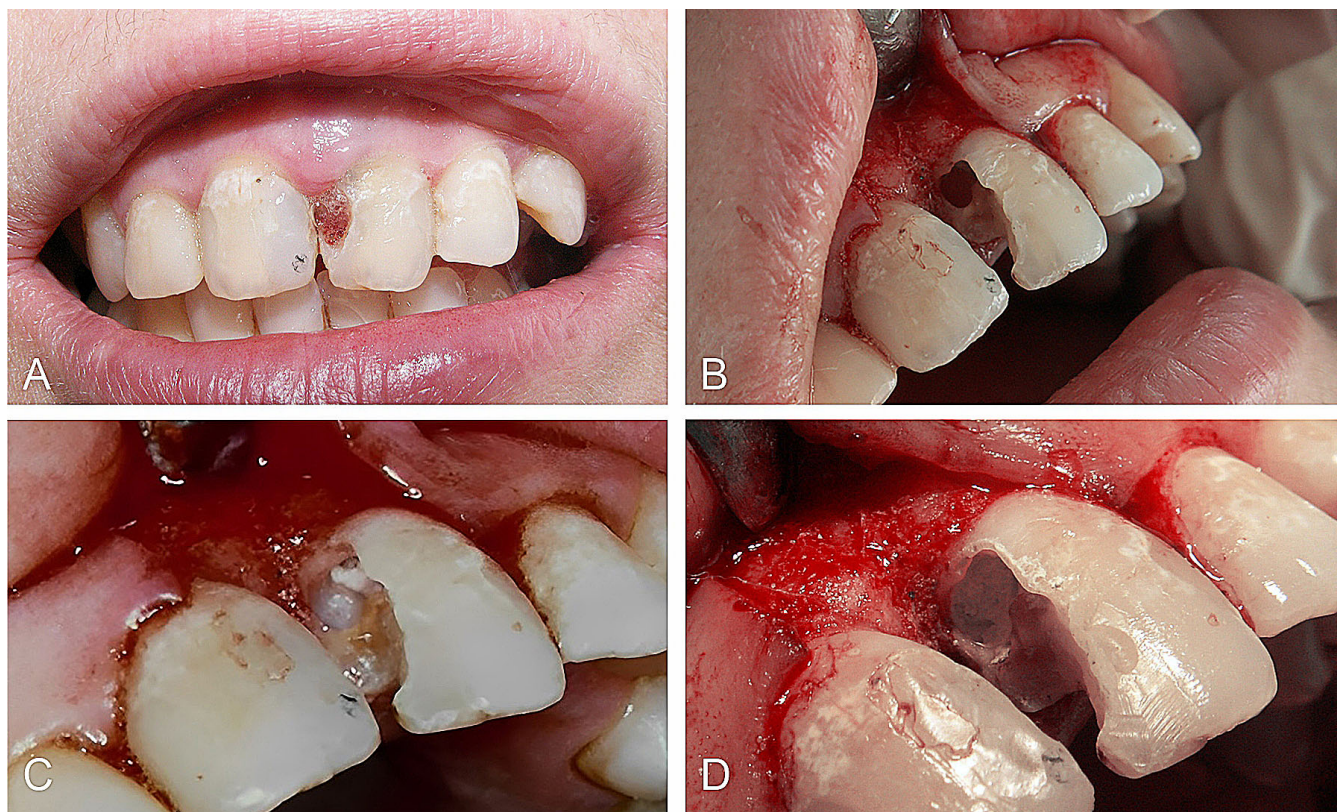
In order to ascertain the three-dimensional characteristics of the lesion, a computed tomography examination was carefully requested, but the patient refused this imaging modality due to its high cost. Due to the case's complexity, an endodontic retreatment of tooth #21 was proposed to the patient along with minor surgery, but she refused the latter. Thus, a conservative treatment approach was adopted. Prior to the root canal retreatment, gingival curettage and root planning were performed.

The subsequent removal of the mesial restoration of tooth #21 revealed a large subgingival cavity complete with granulation tissue. A significant part of the mesial root wall was missing. Initially, an internal approach to debride and seal the defect with mineral trioxide aggregate (MTA) was attempted unsuccessfully (Fig. 1C). Fur-

thermore, despite using the white form of MTA-Angelus (Angelus, Londrina, PR, Brazil), it appeared grayish through the thin labial dentin wall and translucent overlying enamel.

After failing to solve the problem conservatively, the patient gave her informed consent for a surgical treatment approach. Local anesthesia was administered (Ubistesin 40 mg/mL). A small full-thickness buccal mucoperiosteal flap was raised from the maxillary right central incisor to the left lateral incisor (Fig. 2A, B), as well as a palatal flap, limited only to #21. An intrasulcular incision was performed. As the apical margins of the lesion were accessible, no osteotomy was necessary. The granulation tissue was removed from the surgical site using surgical curettes and periodontal Gracey curettes (Hu-Friedy, Leimen, Germany).

Fig. 2. Intraoperative photographs. **A)** Initial facial view. **B)** A full-thickness labial mucoperiosteal flap was raised. **C)** A surgical insight into the uncommon pattern of the lesion. **D)** A white MTA-Angelus, placed into the inner part of the defect.



After the hemorrhage was controlled, the cavity was cleaned and dried. The surgical insight into the lesion revealed a surprising and uncommon picture. Two regular and perfectly smooth concave forms, looking like hemispheres with a bluish color and glossy surface, became visible in the inner part of the defect (Fig. 2C). These hemispheres were partially confluent, as the one near the root canal space was smaller in size. Their appearance significantly differed from that of the surrounding dentin, and when probed, their surface was hard and scratchy. Thus, the initial hypotheses regarding the origin of the lesion were rejected, and the diagnosis of idiopathic cervical resorption was accepted. According to the real

spread of the resorptive defect, it was defined as a Class 3 ICR lesion based on Heithersay's classification [2].

A small cotton pellet dipped in 90% trichloroacetic acid (TCA) was applied in the cavity for 30 seconds. After the use of TCA, the inner defect surface was refreshed with a stainless-steel round bur. Afterward, a "sandwich" technique was used to seal the defect. A white MTA-Angelus was inserted into the root canal space and in the inner part of the lesion (Fig. 1D and Fig. 2D). The mesial root wall and coronal part of the cavity were initially restored with glass ionomer cement (GIC) Ketac Fil Plus (3M ESPE Dental Products, St. Paul, MN, USA) (Fig. 1D and 3A). One week after suture removal, the accessi-

ble supragingival part of the primary restoration was replaced provisionally with the chemically cured composite resin material Compolux (Septodont, Saint-Maur-des-Fossés Cedex, France) (Fig. 3B). Following complete healing of the surgical wound, a definitive restoration was placed on tooth #21 using a light-cured composite resin, Gradia Direct Anterior (GC Corporation, Tokyo, Japan), along with the replacement of the bad-looking restorations on tooth #11 (Fig. 3C).

Short- and middle-term follow-ups (1 to 3 years)

At the regular clinical checkups, the patient was

asymptomatic. The tooth responded normally to percussion and palpation; the gingiva was inflammation-free, but increasing probing depth mesial to the restoration was registered with time. The radiographic follow-ups showed no periapical or periradicular pathology, but a gradual vertical bone loss of the interdental septum between teeth #11 and #21 was observed (Fig. 1E). Moreover, a year after the surgery, a black discoloration appeared in the facial cervical area of the crown (Fig. 3D). Different prosthetic treatment options were proposed to the patient, but she refused any kind of aesthetic rehabilitation. Thus, only periodontal treatment was regularly performed.

Fig. 3. Intraoral facial photographs. **A)** GIC, placed intraoperatively in the outer part of the lesion and in the carious cavity. **B)** Provisional restoration with Compolux. **C)** Definitive restoration of the lesion with Gradia Direct (baseline tooth appearance). **D)** 14-year follow-up, revealing WMTA-induced black cervical discoloration.



Long-term follow-up (14 years)

After the 3-year recall, the patient was not accessible for annual checkups for a long time. Unexpectedly, 14 years after the surgical procedure, she returned to the clinic for a dental checkup without specific complaints. The tooth was symptom-free and functional. With the exception of the dark cervical area, the restorations displayed good quality and color stability (Fig. 3D). A probing depth of 5 mm was measured mesial to the restoration. A slight labial gingival recession was noticeable next to the stained cervical area. Radiographic examination revealed no progression of the resorptive lesion, but significant interdental bone loss was evident (Fig. 1F). The panoramic radiograph of the patient did not disclose any additional pathological findings in the dentition.

At the time of surgery, the patient's medical history was noncontributory. Except for existing restorations, all known local or systemic predisposing factors for cervical resorption were ruled out after a detailed anamnesis, including trauma, bleaching, and orthodontic treatment. And, since the cause of the cervical lesion occurrence was not identified, the condition was referred to as "idiopathic." However, 12 years later, at the age of 43, the patient was diagnosed with a specific genetic disorder, Familial Mediterranean fever (FMF), also known as "Armenian disease." Retrospectively, the patient witnessed that the illness's symptoms had started to show early on, prior to the surgical procedure.

DISCUSSION

Various materials have been reported in the literature for the clinical repair of ICR lesions in the anterior zone—GICs, MTA, or composite resin materials [13-15]. Recently, calcium silicate-based cements (CSCs) have been suggested as the preferred choice due to their excellent properties, including the potential for osseous and periodontal regeneration [3, 14]. However, they exhibit some drawbacks. One limitation associated with CSCs is their potential for discoloration. The use of bismuth oxide as a radiopacifier or blood contamination are two possible causes of tooth crown staining. Although CSCs are considered to possess a low susceptibility to blood contamination, excessive hemorrhage should be minimized by clinicians as it could adversely affect the setting reaction and the biomaterial's properties [14, 16, 17]. Conversely, resin-modified GICs offer an aesthetic appearance and can be polished. However, they exhibit higher cytotoxicity and lower regenerative capacity than CSCs [18].

This case report presents the multidisciplinary surgical management of a Class 3 ICR lesion with an uncommon origin in the upper central incisor. In an effort to gain a good healing outcome along with an aesthetic result, the lesion repair utilized a hybrid technique, combining the excellent sealing ability of MTA in the inner part of the defect with the aesthetic appearance of GIC. Similar treatment approaches have also been applied by other authors [19]. In the case presented herein, however, both materials exhibited their main drawbacks. The loss of epithelial attachment around the restoration was most likely caused by the GIC's lack of regenerative capacity. The cervical crown staining was not avoided even though white MTA-Angelus was used and coated with GIC. ProRoot MTA and Biodentine are the most commonly used CSCs for ICR defect repair in the anterior zone [14, 20, 21]. Gi3n-Guerra et al., however, mentioned the unaesthetic gray translucency of white ProRoot MTA [14]. These findings parallel those reported in this study.

Teeth with ICR fall into a difficult category for achieving a predictable outcome. The prognosis for the treatment outcome of such lesions is uncertain or poor because of their proximity to the critical crestal zone and gingival sulcus [2, 22]. Moreover, the idiopathic etiology of the lesion is believed to contribute to the appearance of recurrences. Long-term clinical follow-up is essential to evaluate the durability and efficacy of various ICR treatments. Due to the rarity of the disease, most published studies are case reports or small case series, and only a few have been followed up for a longer period (2–7 years) [13, 19, 21, 23].

Only two investigations were found to analyze the surgical outcome results based on a homogenous and statistically significant sample of clinical cases. In a noteworthy retrospective study, Jeng et al. assessed the survival and clinical success of 42 ICR-affected teeth after surgical intervention for external repair. The two-year survival rate was 71.20% [24]. Jebril et al. assessed the clinical outcome of a smaller sample of 14 teeth with

ECR, followed up for 20 months on average (8 to 48 months), and reported a survival rate of 100%. However, real clinical success with comprehensive restorative integrity and arrest of the resorptive process was found in only 79% of cases [7].

Several prognostic factors determining the outcome of external cervical resorption management have been discussed in the endodontic literature. According to the authors of two recent retrospective observational studies, the survival rate of repaired ICR lesions appeared to be related to the extent of the defect, regardless of the treatment approach selected [25, 26]. Another recent study suggested that sex, age, and choice of restorative material did not significantly affect the progression of resorption. Patients with solitary lesions had a better outcome than those with multiple teeth affected by external cervical resorption [24]. Regarding the impact of tooth position (posterior or anterior) on the long-term prognosis of ICR treatment, opinions disagree [24, 25].

Notably, an accurate assessment of the true nature of the ECR lesion is important for its appropriate management and successful healing outcome [1, 22]. In the case presented herein, the characteristics of the ICR lesion were very peculiar and uncommon. It had no distinguishing radiographic features but was characterized by a specific color and surface feature in its interior. Initially, the patient's intraoral status and medical history denied all known local and systemic predisposing factors. Only the presence of proximal restorations was recorded.

Interestingly, 12 years after the surgery, the patient was diagnosed with a rare disease known as FMF. FMF is a hereditary autoinflammatory genetic disorder that causes recurrent fever episodes typically accompanied by pain in the abdomen, chest, or joints. It most often occurs in individuals of Mediterranean and Middle Eastern descent, typically Sephardic Jews, Turks, Arabs, and Armenians. The patient belonged to one of these ethnic groups. However, FMF is observed worldwide. FMF attacks usually start in early childhood, and 80%–90% of patients become symptomatic before the age of 20 [27, 28], and that was exactly the case with this patient. Unfortunately, her illness was not diagnosed in a timely manner. The most severe complication of FMF is secondary systemic AA amyloidosis due to the accumulation of extracellular amyloid protein in various tissues due to chronic inflammation. It clinically manifests as progressive nephropathy, leading to end-stage renal disease [28].

Over the last few years, significant progress has been observed in research on the pathogenesis, genetic testing, diagnosis, comorbidities, disease-related damage, and treatment approaches to FMF. Elucidation of some pathogenic mechanisms has led to the discovery of pathways involved in inflammatory, metabolic, cardiovascular, and degenerative diseases [27]. Recent publications have described its frequent association with other diseases and/or syndromes, the common denominators of

which are genetic predisposition, immune dysfunction, and autoinflammation, such as spondyloarthropathies, systemic vasculitis, and inflammatory bowel disease [29].

However, the connection between this disease and the occurrence of ICR remains uncertain. Most commonly, external root resorption affects multiple teeth when associated with systemic disorders [10-12]. The lesion in the clinical case reported herein was isolated. On the other hand, ICR is considered a multifactorial disease [1, 22]. Whether FMF was related to the initiation of the ICR or only contributed to it, this question remains open, as no such case has been documented so far. The hypothesized impact of FMF on ICR development requires further interdisciplinary investigation.

CONCLUSIONS

The so-called “idiopathic” ICR lesion may exhibit unusual appearance and features. Clinicians should be alert for the occurrence of such a lesion, especially in patients with concomitant systemic disorders. The right choice of appropriate repair material is a key factor in the healing outcome of ICR lesions in the anterior zone.

Abbreviations:

CSCs - calcium-silicate cements
FMF - Familial Mediterranean fever
GICs – glass ionomer cements
ICR – invasive cervical resorption
MTA – Mineral trioxide aggregate
TCA - trichloroacetic acid

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