

Short Case Report

Impacted canine and ossifying fibroma: case report and literature review

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Abstract – Introduction: An impacted tooth is a permanent tooth that has a completely developed root but which has failed to erupt. Many etiologies can lead to impaction, including a mechanical obstacle, such as benign tumor. **Observation:** An 11-year-old girl was referred for the avulsion of the left mandibular permanent canine, which was impacted. A radiolucent lesion with ground glass opacity was found on the eruption pathway of the tooth. Anterior radiographs showed the lesion that seemed to be the cause of the impacted canine. Anatomopathological examination revealed the lesion to be a juvenile trabecular ossifying fibroma. **Comments:** Ossifying fibromas are rare jaw tumors. They can be of various types: ossifying fibroma, psammomatoid, or trabecular juvenile ossifying fibroma. This is the first case that clearly shows the association between ossifying fibroma and tooth impaction. A literature review of ossifying fibromas and the management of fibro-osseous benign lesions of the jaws was performed. **Conclusion:** Although benign, juvenile trabecular ossifying fibroma can be locally aggressive and has a high rate of recurrence. It is very important to establish a precise diagnosis of a fibro-osseous benign lesion of the jaws to plan optimal management.

Introduction

An impacted tooth is a definitive tooth whose root has formed completely but whose crown has not erupted [1]. The causes of impacted teeth, among others hereditary factors, include dysfunction of some endocrine glands; mechanical obstacles such as vestibuloversion of the mandibular incisors, fracture of the mandible near the site of canine eruption, premature loss of deciduous canines, or abnormal shape of adjacent teeth; and/or tumoral pathologies such as odontomas or cysts [1–4].

Ossifying fibromas (OF) are benign tumors that mainly affect the bones of the face. They are a type of benign facial fibro-osseous lesions (FOBLs) [5]. According to Brannon *et al.* (2001), these lesions are divided into three categories according to their origin: neoplastic lesion (OF), dysplastic lesion, the origin of which would probably be the alveolar-dental ligament (bone dysplasia), and developmental lesion (fibrous dysplasia) [6,7]. Ossifying fibromas can be of three

types: ossifying fibromas (OF), juvenile trabecular ossifying fibromas (JTof), or juvenile psammomatoid ossifying fibromas (JPOF).

This article analyzes the case of an impacted mandibular canine because of its association with JTof along with a radiological follow-up over 4 years.

Clinical case

An 11-year-old girl was referred by the Orthodontic Department, where she had been followed up for 3 years for a dentomaxillary disharmony, to extract the impacted left mandibular canine. Her medical history included hexadactyly, precocious puberty in the context of sustained obesity, treatment with triptorelin 3.75 mg (Gonapeptyl LP® 3.75 mg every 4 weeks), and multiple fractures (left foot and right ankle).

On orthopantomography (OPT), the left mandibular canine was almost horizontal, and its crown was superimposed on the roots of the mandibular incisors, with a poorly defined radiolucent image surrounding the crown and extending distally between the lateral incisor and the first left mandibular

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Fig. 1. OPT showing the impacted canine and the radiolucent lesion.

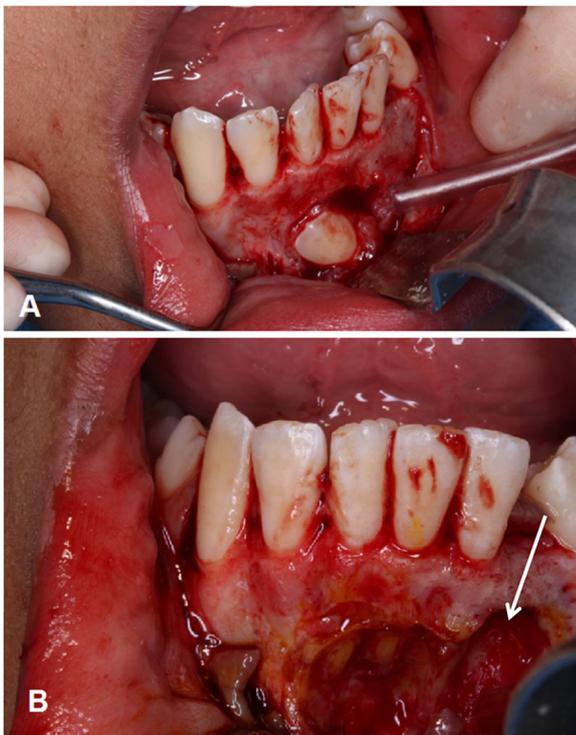


Fig. 2. (A) Intraoral photograph during the intervention showing the left mandibular canine position. (B) Intraoral photograph showing the closeness of the root apex to the canine. The arrow indicates the bony cleavage plane of the lesion.

premolar (Fig. 1). Examination of previous orthopantomograms showed that the lesion had always been present and had changed little over 3 years. The left mandibular canine already had a more mesial position and a more pronounced coronomesial version than the contralateral mandibular canine 3 years ago. These abnormalities became more pronounced over time, probably because the lesion was an obstacle to the normal eruption of the tooth (Fig. 2).

Cone beam computed tomography was performed to determine the association between the tooth and other adjacent anatomical structures (Fig. 3). The tooth was placed buccally to the mandibular incisors. The radiolucent image showed two aspects: A radiolucent lesion at the canine crown and ground glass opacity; the margins were clear, and there was no damage to adjacent anatomical structures.

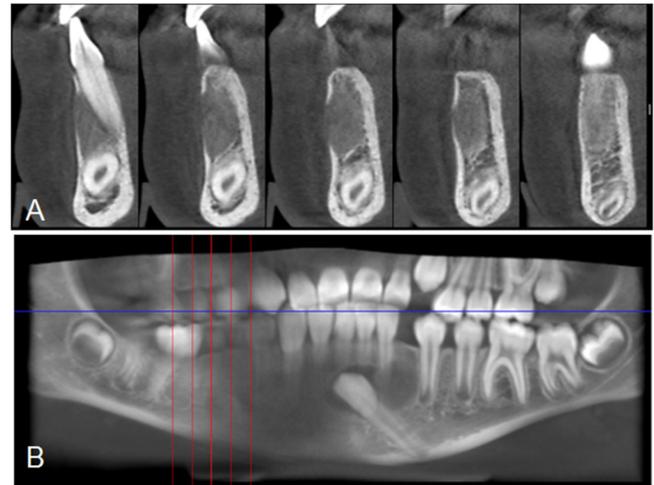


Fig. 3. (A) Axial section showing the radiolucent lesion between 32 and 34. (B) Reconstruction showing the proximity of the tooth crown to the mandibular incisors root apex. Two aspects of the lesion can be seen: the part enclosing the tooth crown appearing radiolucent and a distal extension with ground glass opacity.

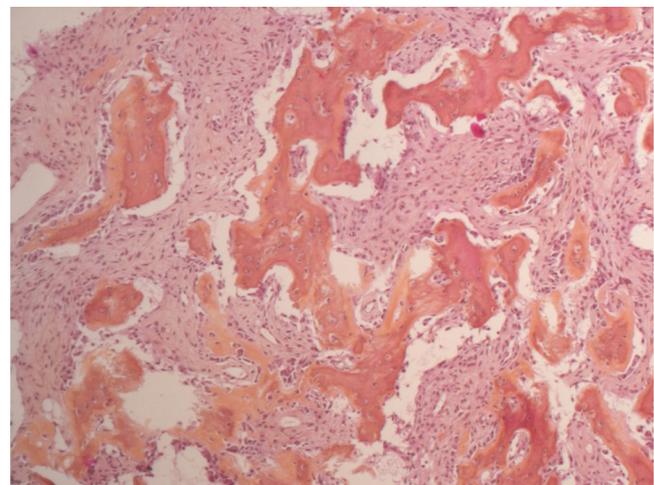


Fig. 4. Histological section showing osteoid trabeculae (H&E, $\times 40$).

Orthodontic treatment of interception included the extraction of the temporary teeth and the installation of a space maintainer (lingual arch) to allow its eruption, which never occurred. Orthodontic surgical traction carried a great risk of causing root lesions on the mandibular incisors. Thus, it was decided to extract the canine under local anesthesia. This was part of an overall treatment plan for the extraction of the maxillary second premolars and the right second mandibular premolar.

Once the tooth was extracted, an exploration of the distal lesion was performed; it showed a wet sandy appearance and was well defined; curettage was easy with a well-defined cleavage plane (Fig. 4). Following curettage, a macroscopically healthy bone was found, with a well-individualized cortex.

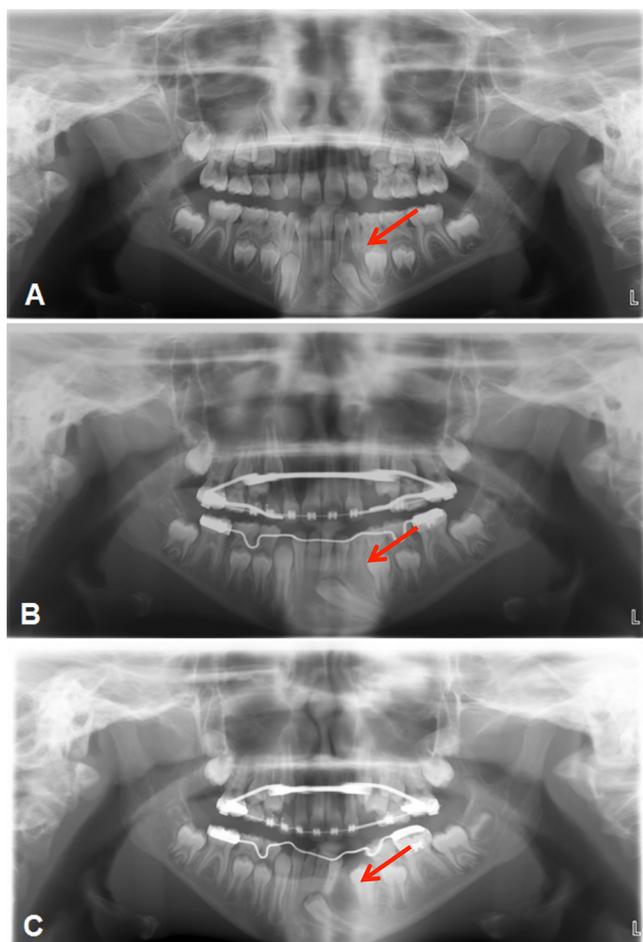


Fig. 5. OPT taken 3 (A), 2 (B), and 1.5 (C) years before surgery.

Anatomopathological examination showed neoformed masses and anastomosed bones sometimes surrounded by osteoblasts, which were separated by a moderately cellular fibrous tissue, leading to the diagnosis of OF (Fig. 5). OFs can be of three different types, and postoperative monitoring differs according to the type. Although slow lesion growth is a confounding factor, JTOFs grow rapidly and are sometimes aggressive. In consultation with the pathology team, after rereading the sections, and based on clinical presentation (slow growth, tooth displacement, unencapsulated lesion, and osteoid trabeculae), the diagnosis of JTOF was confirmed.

Eighteen months after the procedure, the patient showed no signs of a recurrence. A 5-year follow-up was instituted to monitor recurrence.

Discussion

BFOL of the craniofacial region can form several types of lesions that are very similar to each other from a histological, clinical, and radiological point of view: fibrous dysplasias, bone dysplasias, and OF. These pathologies result in the replacement of the normal bone of the fibrous connective tissue, forming varying amounts of osteoid, bone, or cement-like calcifications [5,6].

These three types of lesions, which are histologically similar, are differentiated based on clinical, radiographic, and histological findings [6,7]. Patients' age, their ethnic origin, and the sites affected by the lesion are important for establishing the diagnosis [6,8].

In the present case, anatomopathological examination indicated OF, but a specific type was not determined. In fact, there are three types of OFs whose different clinical and radiological characteristics are described in Table I.

OF usually occurs between the age of 20–30 years. Women are more frequently affected than men, as are Caucasian patients, followed by patients of African descent. The posterior mandibular regions are the most affected sites.

The origin of OFs is not unanimous; while the World Health Organization considers it to be a fibro-osseous neoplasm, others consider it to be an odontogenic tumor [5,9]. Often, it evolves aggressively but slowly, is asymptomatic, and is discovered accidentally. Its size at the time of discovery varies greatly (1–5 cm) [6,10]. Typically, the adjacent structures move as the lesion grows (teeth and inferior alveolar nerve). Radiologically, it presents as a unilocular lesion, well demarcated from the surrounding bone, in majority of the cases. A fibrous capsule around the lesion is usually found during excision [6,8,11].

Juvenile OFs (JOFs) are fibro-osseous tumors that are rarer and more aggressive than OF. The distinction with JOF and OF is sometimes challenging because of their clinical, radiological, and microscopic similarities [7,12]. They exhibit rapid growth, sometimes growing to large sizes. Radiologically, JOF is also well delimited, but unlike OF, it lacks radioclear peripheral borders (non-encapsulated tumors) [6,8].

There are two histological types of JOF: JPOF and JTOF. JPOF essentially affects the bones of the orbits and the paranasal sinuses, and the pathological aspect is not similar to that found in the presented case (presence of fusiform fibroblast cells and psammomatoid calcifications) [6]. JTOF most often affects children between 8 and 12 years of age, with a male predominance. The maxillary bone is the most frequently affected site [6,9]. Microscopically, it is rich in fibrous tissues and osteoid deposits and shows trabecular bone surrounded by rounded osteoblasts.

In the present case, the lesion grew slowly, did not involve any encapsulation, and showed osteoid trabeculations on anatomopathological examination. Thus, the diagnosis of JTOF was made, although it did not evolve aggressively similar to most lesions. Similarly, Brannon *et al.* (2001) have reported that this type of lesion does not always behave aggressively, which is consistent with the present report [7].

OF treatment involves curettage in the context of small lesions and radical excision (marginal or segmental resection) in case of very large, debilitating, or rapidly evolving lesions [14–16]. These lesions show a very high rate of recurrence (30%–58%) [13,14,16]. Clinical and radiological monitoring is essential.

Although one case of an impacted canine resulting from OF has already been reported in the literature [17], this case is the

Table I. Comparison of different types of ossifying fibromas, after Zegali *et al.* (2015) [13].

	Cemento-ossifying fibromas	Trabecular juvenile ossifying fibromas	Psammomatoid juvenile ossifying fibromas
Age	20–40 years	From 15 years	20–30 years
Location	Mandible, dental area	Mandible, non-dental areas	Craniofacial bone, paranasal sinus
Clinical signs	Slow growth	Slow to very fast growth, facial deformity	Slow growth in periorbital areas
Radiology	Well demarcated, radiodensity varies according to the age of the lesion Dental displacement with minor root resorption	Well defined, single or multi-celled Variable radiodensity Root resorption and dental displacement	Well defined, radiolucent Obturation of the nasal and sinus cavities
Histology	Variable, encapsulated	Well demarcated, non-encapsulated, osteoid trabeculae	Well demarcated, non-encapsulated, psammomatoid appearance
Recurrence	Rare	Frequent (30%–50%)	Frequent (30%–56%)

first to show the direct cause–effect relationship between an impacted canine and OF.

It is very important to make a precise diagnosis of different FOBLs. Although they are histopathologically similar, their evolution and management varies greatly depending on the pathology. All OFs require resection because of their neoplastic nature [6,8]. OD, including periapical bone dysplasia, focal bone dysplasia, and florid bone dysplasia, does not require treatment; however, annual monitoring is essential. In fact, periapical and focal bone dysplasias may progress to florid oral bone dysplasia, which may undergo sarcomatous transformation in very rare cases [6].

In the case of fibrous dysplasia affecting the jaws, the treatment approach should be modulated according to patient age, lesion size, and lesion growth rate. Often, aesthetic concerns determine the course of action, ranging from simple clinical and radiological monitoring to osteotomies for modifying bone surfaces. Long-term monitoring is necessary because of the risk of sarcomatous transformation [8,18].

Conclusion

This article details the case of an impacted canine related to JTOF that was present for several years. The diagnosis of these lesions is difficult because of their clinical, radiological, histological similarities with other FOBLs or OFs. The establishment of a precise diagnosis is essential to determine the course of action, which varies greatly depending on the type of lesion. These are rare, frequently recurrent lesions; thus, close monitoring should be performed over several years.

Conflicts of interests: The authors declare that they have no conflicts of interest in relation to this article.

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