Journal of Cranio-Maxillo-Facial Surgery 41 (2013) 391–396

Contents lists available at SciVerse ScienceDirect

Journal of Cranio-Maxillo-Facial Surgery

journal homepage: www.jcmfs.com

Case report

Atypical presentations of simple bone cysts of the mandible: A case series and review of literature

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ARTICLE INFO

Article history:
Paper received 15 May 2012
Accepted 5 November 2012

Keywords:
Intraosseous radiolucencies
Simple bone cyst
Atypical
Mandible

ABSTRACT

Simple bone cysts are well-defined intraosseous radiolucencies that often extend between the roots and appear clinically like empty cavities. This article aims to provide more information about this lesion with limited prominence in academic literature, to illustrate atypical cases, and to provide a review of the current literature. A series of six atypical cases of simple bone cysts is presented and their clinical, radiographic and microscopic characteristics, differential diagnosis, treatment and follow-up are discussed. Correct diagnosis of this entity is of key importance, since it presents with clinical & radiographic similarities to other bone lesions, some exhibiting more aggressive behaviour.

1. Introduction

Simple bone cysts (SBCs) of the jaw were initially described as an entity in 1926 (Lucas and Blum, 1929), yet their diagnostic criteria were only established in 1946 (Rushton, 1946). SBCs are classically described as well-defined radiolucent lesions, often with scalloping of the margins between the roots of standing teeth. Clinically, these lesions demonstrate an empty cavity devoid of an epithelial lining, which are eventually filled with serous or sanguinous fluid (Rushton, 1946; Jundt, 2005). SBCs have been described using different terms in the literature, such as traumatic bone cysts, haemorrhagic bone cysts, extravasation cysts, solitary bone cysts, idiopathic bone cysts, idiopathic bone cavities and progressive bone cavities (Suei et al., 2007; Cortell-Ballester et al., 2009).

This intraosseous lesion is not unique to the jaw and is more commonly found in long bones (90% of cases), mainly affecting the metaphyseal region of the proximal ends of the humeral (65%) and femoral shafts (25%) (Lokiec and Wientroub, 1998). SBCs of the jaw bone appear to be far less frequent (1%), and the body of the mandible is usually affected (75%), mainly in the pre-molar and molar regions (Copete et al., 1998). Maxillary lesions may also occur, but these are considered exceptionally uncommon (Mitchell and Ward-Booth, 1984; Kuhmichel and Bouloux, 2010).

SBCs are most often diagnosed in the first 2 decades of life, with a slight female predominance, and are rarely seen after 25 years of age (Tong et al., 2003). One important aspect is the fact that SBCs are usually asymptomatic, do not tend to cause bone swelling and their diagnosis often results from routine radiographic examination. The radiographic pattern is described as a solitary, well-circumscribed radiolucent image with scalloped or irregular margins, which are well-defined. Rarely, expansion of the cortical bone is noticed (Forssell et al., 1988), and although highly suggestive, these image features are not pathognomonic (Suomalainen et al., 2009). Even though the diagnostic criteria and radiographic features of SBC were previously defined, they do not always appear in a classic pattern. According to a few publications, the variations of SBCs can include a multilocular appearance, an association with impacted teeth, multiple occurrences in the same patient, asynchronous, and unusual sites (Forssell et al., 1988; Tong et al., 2003; de Souza Noronha et al., 2012).

Taking these considerations into account, SBCs could mimic other radiographic bone lesions, including those with more aggressive behaviour, such as ameloblastoma, keratocystic odontogenic tumour, myxoma, and central giant cell granuloma (Pitak-Arnnop et al., 2010; Shirani et al., 2011; Kansy et al., 2012).

Complementary examination, such as pulp vitality testing, computed tomography (CT) and biopsy, is therefore important for establishing a proper diagnosis. Differential diagnoses include keratocystic odontogenic tumours, central giant cell lesion, ameloblastomas, myxomas and other odontogenic cysts and tumours.

Apparently, the association of the surgical finding of an empty cavity with the above clinical and radiographic characteristics is

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http://dx.doi.org/10.1016/j.jcms.2012.11.002

enough to establish the diagnosis of an SBC (Homem de Carvalho et al., 2010). However, when material can be obtained during the surgical procedure, histopathological analysis is mandatory and usually diagnostic.

As a result, the aim of this study is to report the details of clinical, radiographical and surgical findings, as well as the outcome and long term follow-up of six unusual SBCs affecting the mandible.

2. Case reports

2.1. Case 1

A 16-year-old boy with temporomandibular joint disorder was referred to an orthodontist for treatment. On radiographic examination, a multilocular radiolucent region involving the left posterior body and ascending ramus of the mandible was found. The patient was then referred to our clinic (Fig. 1A). Extension of the lesion with marginal scalloping was noticed into the inter-radicular regions of the lower left molars. Even though the first, second and third molars were involved, they were vital. The third molar was impacted in a mesioangular position, but the lesion had been asymptomatic. Computer tomography revealed smooth bone expansion and signs of perforation of the lingual cortex (Fig. 1B). With the provisional diagnosis of an aneurysmal bone cyst or ameloblastoma, a fine needle aspiration was performed, but no material was obtained. As a result, a surgical exploration was executed and, after raising a mucoperiosteal flap, an empty cavity was found with the inferior alveolar nerve lying in the middle of the empty cavity. Microscopic examination of the overlying cortical bone showed small amounts of granulation tissue and no signs of epithelial lining, with fibrous material attached to the cortical bone. The final diagnosis was of SBC. Radiographic examination 3 months after the biopsy showed reduction in size of the cavity, and evidence of total bone healing was detected at the 1 year follow-up appointment (Fig. 1C).

2.2. Case 2

A 19-year-old girl was referred to our clinic by her orthodontist who had noticed the presence of a unilocular radiolucency on radiographic examination. The radiolucent area had scalloped, well-circumscribed margins involving the left ascending ramus of the mandible and a relationship to the third molar (Fig. 2A). No evidence of caries was seen, and the tooth was viable. No medical history was considered relevant. Differential diagnoses included an ameloblastoma, odontogenic keratocyst or giant cell lesion. The third molar was extracted and surgical exploration of the radiolucent area in the left mandibular ascending ramus detected an empty bone cavity that was carefully curetted around the cavity walls. The bone tissue related to the lesion was taken for microscopic examination and showed normal cortical bone, fragments of osteoid tissue and no signs of epithelial tissue. The final diagnosis made was that of a simple bone cyst. The patient then defaulted on follow-up and was only located 2 years later. Radiographic examination showed that the lesion had completely resolved (Fig. 2B).

2.3. Case 3

A 15-year-old girl was referred by her orthodontist to our clinic for the investigation of a multilocular radiolucency extending from the right ascending ramus. The lesion had passed through the angle and body of the mandible and had reached the canine region, with margins extending between the roots of associated teeth in a scalloped and corticated pattern (Fig. 4A). There was a mild facial asymmetry on the right side (Fig. 3A). Intraorally, a slight bone swelling of the lingual and labial cortices was noticed (Fig. 3B). CT images confirmed expansion of the buccal and lingual cortex of the right ramus and body of the mandible (Fig. 4B). The lesion was asymptomatic and was found incidentally on radiographic examination. There was no history suggestive of any underlying medical problem and all related teeth were viable. After the provisional diagnosis of an ameloblastoma, odontogenic keratocyst or central giant cell lesion, a needle aspiration was performed and was shown to be negative for any material. Subsequently a surgical exploration was performed of the involved region, revealing an empty bony cavity without any sign of cyst lining or capsule (Fig. 3C). The material taken was submitted for histopathology. Analysis report was compatible with SBC (Fig. 3D). After 4 months of follow-up, a CT examination showed an increase in the degree of bone density of the lesion in the molar region compared with the

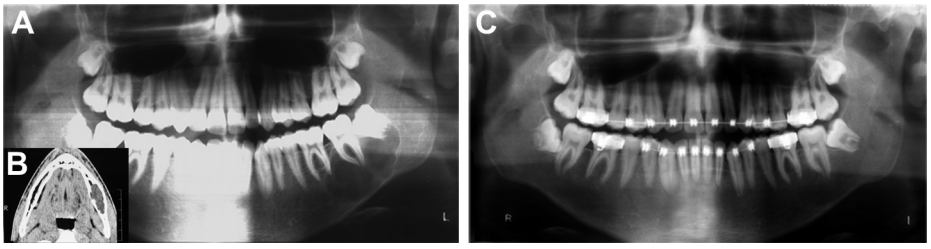

Fig. 1. A. Panoramic radiograph showing a large radiolucent, multilocular lesion of the left posterior mandibular body with scalloped margins. **B**. CT, revealing smooth bone expansion and erosion of lingual cortex. **C**. Panoramic radiograph 1 year post-operative demonstrating complete ossification of the defect.

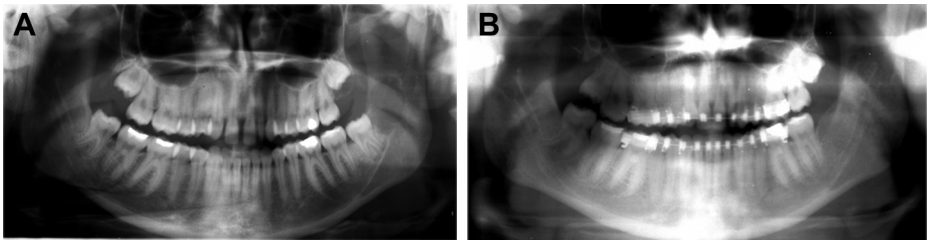

Fig. 2. A. Panoramic radiograph revealing a unilocular radiolucent, with scalloped, well-circumscribed margins involving the left ascending ramus of the mandible with relationship to the third molar. **B**. Panoramic radiograph 2 years post-operative showing complete bone healing.