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Associated aneurysmal bone cyst and cemento-osseous dysplasia: a case report and review of the literature

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The purpose of this case report is to describe a previously unpublished association between focal cemento-osseous dysplasia (FCOD) and an aneurysmal bone cyst (ABC) and review the literature with regard to associated benign fibro-osseous lesions and cysts. A 41-year-old woman without a history of trauma presented with asymptomatic swelling in the right side of the mandible. Radiographs of the region revealed a unilocular radiolucent area with radiopaque foci. After aspiration of the lesion was positive for serosanguineous fluid, complete excision of the lesion was performed. Microscopic examination revealed a hybrid ABC and FCOD. The 12-month follow-up showed significant bone repair and no signs of recurrence. A review of the English-language literature from 1980 to 2012 revealed 1 retrospective study, 4 case series, and 18 single-case reports on the topic of cemento-osseous dysplasias, fibro-osseous lesions, and aneurysmal bone cysts. Of 59 cases, none reported an association between an ABC and FCOD. Although fibro-osseous lesions do not require intervention, surgical excision is recommended when they are associated with cysts. This case, in which an ABC and FCOD were associated, reinforces the need for a careful diagnostic process in radiographically mixed lesions that respond positively to aspiration biopsy.

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Key words: aneurysmal bone cyst, fibro-osseous lesions, focal cemento-osseous dysplasia, hybrid lesions

An aneurysmal bone cyst (ABC) is a benign lesion that may behave in a locally aggressive manner, demonstrating rapid growth and osteolytic potential. Its pathogenesis is related to alterations in vascular homeostatic balance within the bone and an increase in venous pressure.[1-3] ABCs commonly present as radiolucent, expansive lesions with well-defined margins and destruction of cortical bone, which may or may not be accompanied by painful symptoms.

ABCs may be classified as primary or secondary lesions.[1,4] When no subjacent pathosis is identified, the ABC is considered a primary lesion, its most frequent form (70%).[2,3] When superimposed on a preexisting bone lesion, it is classified as a secondary lesion.[2,3] There are reports of concomitant presentation

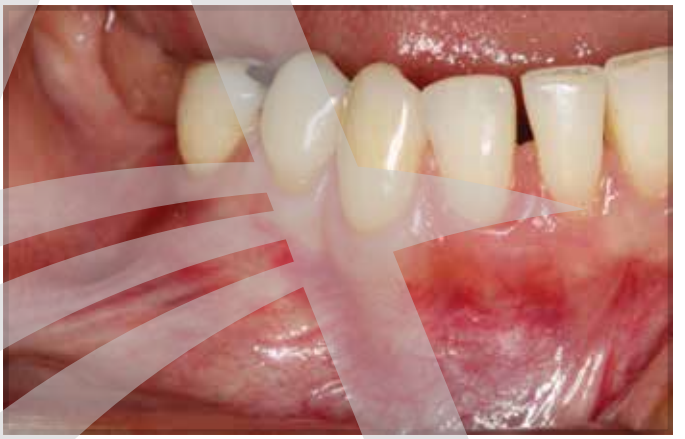

Fig 1. Initial presentation of a lesion involving the mandibular premolars. There is notable vestibular cortical expansion.

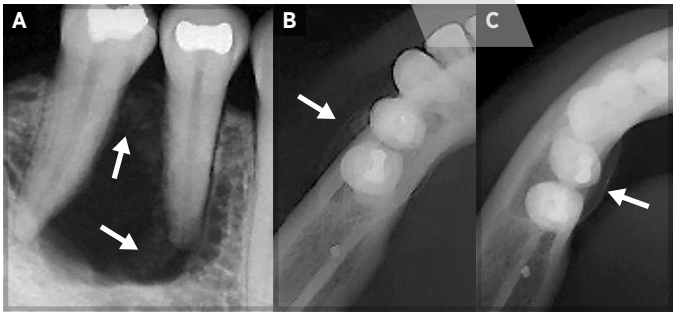

Fig 2. Initial radiographic presentation of the lesion. A. Mineralized tissue in the distal region of the mandibular first premolar (arrows). B. Vestibular cortical expansion (arrow). C. Lingual cortical expansion (arrow).

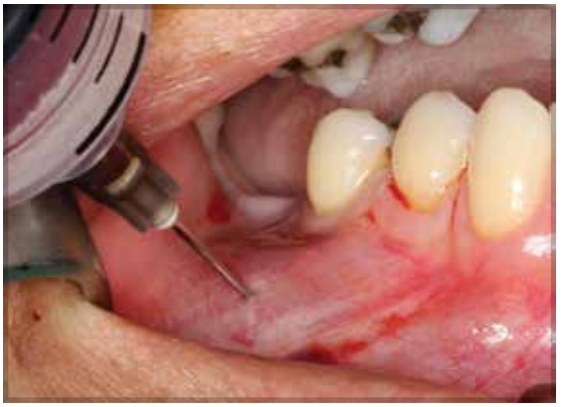

Fig 3. Aspiration puncture positive for presence of serosanguineous fluid.

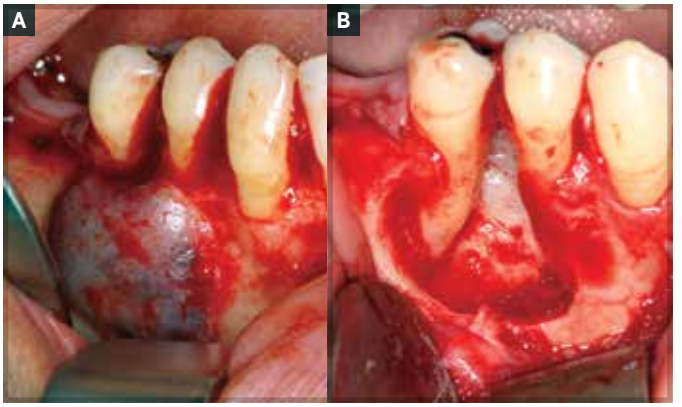

Fig 4. Enucleation of the lesion. A. Exposure of the lesion. B. Surgical bed immediately after enucleation. The root apices of the premolars were in intimate contact with the lesion.

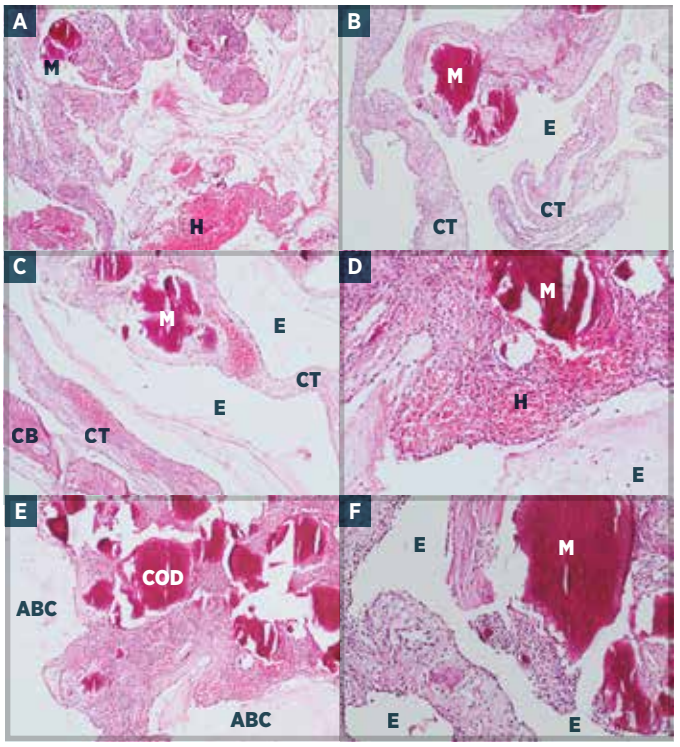

Fig 5. Microscopic aspects of the hybrid lesion (hematoxylin and eosin stain; Fig 5A, 5B, 5C, 5E, original magnification 4×; Fig 5D, 5F, original magnification 10×). ABC, aneurysmal bone cyst; CB, cortical bone; COD, cemento-osseous dysplasia; CT, strips of connective tissue with capsular aspect; E, fibrinohemorrhagic exudate without endothelium; H, hemorrhagic areas; M, mineralized tissue.

of fibro-osseous lesions and ABCs; however, in the oral and maxillofacial region, only fibrous dysplasia and ossifying fibromas have been mentioned.[5-14]

Cemento-osseous dysplasia (COD) is the disorderly production and development of bone and material similar to cement.[3] CODs may be related to a hormone imbalance that influences bone remodeling, justifying a greater prevalence in women. There are 3 basic subtype variants of the same process: focal (association with the apices of teeth or in areas of previous tooth extraction in the posterior mandibular region), periapical (generally found intimately associated with mandibular anterior teeth), and florid (involving at least 2 quadrants of the maxillae) cemento-osseous dysplasia.[15]

Clinicians may have difficulty in identifying the clinical and radiographic characteristics of these lesions, which leads to erroneous diagnoses and inadequate procedures. This case report will relate and discuss a previously unreported association between an ABC and focal cemento-osseous dysplasia (FCOD) and review the literature on possible relationships between fibro-osseous and cystic lesions.

Case report

The patient, a systemically healthy 41-year-old woman of African-Brazilian descent, presented to the Stomatology Clinic of the State University of Maringá, Brazil, with a firm, asymptomatic tumefaction, without inflammatory signs, between her mandibular right premolars (Fig 1). The swelling had been present for 2 months, according to the patient's report. Panoramic, occlusal, and periapical radiographs demonstrated a unilocular, radiolucent area, with small radiopaque foci and well-defined margins, involving the region of the premolar teeth (Fig 2). Slight lingual cortical expansion, light apical resorption of the first premolar, and root displacement of the involved teeth were observed. Both premolars presented pulpal vitality and absence of mobility. The patient denied any history of previous local trauma.

Excisional biopsy was performed under local anesthesia after aspiration puncture was positive for the presence of serosanguineous fluid (Fig 3). The lesion was easily detached from the bone (Fig 4). The apices of the adjacent premolar were carefully submitted to curettage, and the mucoperiosteal flap was replaced and sutured with 3-0 monofilament nylon thread.

In the histopathologic analysis, large hemorrhagic areas and long strips of connective tissue with a capsular aspect were observed (Fig 5A & 5B). In addition, although there were focally dilated spaces containing fibrinohemorrhagic exudate and demonstrating a vascular aspect (Fig 5C), no endothelium was visible (Fig 5D), which helps to differentiate ABCs from intraosseous hemangiomas. Numerous mineralized structures with

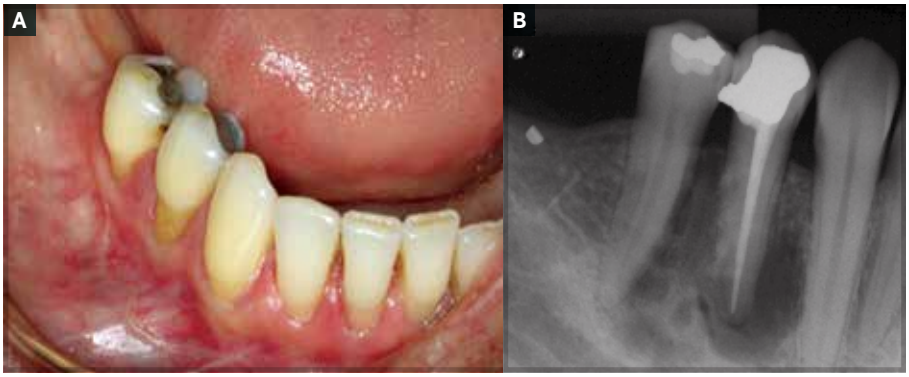

Fig 6. View at 12-month postoperative recall. A. Clinically, there is no sign of recurrence. B. Bone repair is in progress.

different sizes and degrees of maturation were also present, permeating a cellular and well-vascularized connective tissue (Fig 5E). These structures showed a tendency to coalesce in a globular arrangement and presented a cementoid aspect, configuring an FCOD (Fig 5F). Cellularity was not as prominent as would be the case in neoplasms, such as cemento-ossifying fibroma, which would exhibit poorer vascularity. The tendency to coalesce is also a feature observed in cemento-osseous dysplasias, which is useful to differentiate it from other benign fibro-osseous lesions. The concomitant presentation of these histopathologic characteristics was compatible with a diagnosis of an ABC associated with COD, corroborating the radiographic images and biological behavior.

Endodontic treatment was performed in the first premolar 1 month after the biopsy, because, during the biopsy, the lesion was shown to be intimately associated with the tooth and, after the intervention, the premolar no longer responded positively to the pulpal vitality test. This treatment was intended to prevent development of an apical lesion and root resorption. At the 12-month follow-up, bone repair was evident at the site, and there were no signs of recurrence (Fig 6).

Discussion

A search was conducted in the MEDLINE and LILACS databases, using the terms (*cemento-osseous dysplasia*) OR (*fibro-osseous lesions*) AND (*cyst*) OR (*aneurysmal bone cyst*). A search of articles published from 1980 to 2012 retrieved a total of 59 clinical cases in which there was association between cystic lesions and benign fibro-osseous lesions (Table).[5-14,16-26] The publications included 18 single-case reports, 4 case series, and 1 retrospective study.

The most common association related in the literature was that of a simple bone cyst and COD, totaling 41 cases; the next most frequent associations were the concomitant appearance of an ABC and fibrous dysplasia, with 8 cases, and an ABC and an ossifying fibroma, with 7 cases, including the psammomatoid and trabecular juvenile variants.[5-14,16-25] Two cases of simple bone cysts were also found in association with ossifying fibroma and with fibrous dysplasia.[22,26] Sanjai et al related the case of an association between COD and a dentigerous cyst.[23] No association of COD and an ABC was reported in the literature, making the current case presentation unprecedented.

According to Wojno & McCarthy, the concomitant presentation of ABCs and benign fibro-osseous lesions is known in extracranial bones but is infrequent in the craniofacial bones.[7] They reported that 30% of ABCs occur in association with preexisting lesions. According to the literature consulted, their most common association is with ossifying fibroma, followed by fibrous dysplasia.[5-13] The variety of associations with ABCs makes some authors consider these lesions to be hybrids.[6,11] According to Saheeb et al, it is necessary to perform serial microscopic cuts of the entire specimen in order to detect coexistent lesions.[8]

In the majority of cases related in the literature, CODs are associated with simple bone cysts.[16-25] Alsufyani & Lam conducted a study of the clinicoradiographic and demographic characteristics of CODs in a Canadian population.[27] The majority of cases (78.8%) involved the periapical subtype, while 21.2% constituted the florid variant. No case of FCOD was found. In 12.7% of the sample, the lesion was associated with 1 or more simple bone cysts. The authors suggested that this association could make diagnosis difficult.[27]

Chadwick et al related that, in 87% of cases, this association occurs in women in the fifth decade of life, whereas simple bone cysts have no predilection for gender and occur in adolescents.[24] Moreover, the authors affirmed that when both lesions are associated, there is a greater trend toward expansion and thinning of the cortical bones and the formation of "scalloping" between the teeth involved. They found no association of the simple bone cyst with periapical CODs.[24] Cystic lesions were exclusively in the regions of the mandibular molars and premolars. The authors postulated that these lesions are related to a disturbance in cell differentiation (eg, of the osteoblasts and osteoclasts) during growth and ossification of the mandible. They also hypothesized that solitary simple bone cysts may be the same lesion as cysts that are in association with CODs but may "arise owing to different biological circumstances."[24]

Alawi stated that large fibro-osseous lesions may undergo cystic degeneration, leading to the development of simple or aneurysmal bone cysts.[15] The development of an ABC subjacent to a preexisting benign fibro-osseous lesion is suspected when a lesion exhibits rapid growth after years of indolent biological behavior.[3,7] Cysts form as a result of posthemorrhagic osteolysis caused by an increase in venous pressure induced by the lesion.[28]

Table. Previously reported associations between cystic lesions and benign fibro-osseous lesions.

Authors (year)	Study design	Sample size	Association
El Deeb et al (1980)[5]	CR	1	FD & ABC
Blayney & el Tayeb (1986)[6]	CR	1	OF & ABC
Higuchi et al (1988)[16]	CS	4	COD & SBC
Wojno & McCarthy (1994)[7]	CS	5	FD & ABC
Miyauchi et al (1995)[17]	CR	1	COD & SBC
Wakasa et al (2002)[18]	CR	1	COD & SBC
Mahomed et al (2005)[19]	CS	7	COD & SBC
Mupparapu et al (2005)[20]	CR	1	COD & SBC
Saheeb et al (2007)[8]	CR	1	OF & ABC
Składzieriń et al (2008)[9]	CR	1	FD & ABC
Mupparapu et al (2008)[21]	CR	1	COD & SBC
Nasser (2009)[10]	CR	1	POF & ABC
Zillo Martini et al (2010)[22]	CS	2	COD & SBC
Zillo Martini et al (2010)[22]	CR	1	FD & SBC
Sanjai et al (2010)[23]	CR	1	COD & DC
Chadwick et al (2011)[24]	R	23	COD & SBC
Rao et al (2011)[25]	CR	1	COD & SBC
Sankaranarayanan et al (2011)[11]	CR	1	JOF & ABC
Silva et al (2011)[12]	CR	1	TJOF & ABC
Geraldo et al (2012)[13]	CR	1	FD & ABC
Geraldo et al (2012)[13]	CR	1	JPOF & ABC
Gnanadeepam & Ponniah (2012)[26]	CR	1	OF & SBC
Tolentino et al (2012)[14]	CR	1	JPOF & ABC

Abbreviations: ABC, aneurysmal bone cyst; COD, cemento-osseous dysplasia; CR, case report; CS, case series; DC, dentigerous cyst; FD, fibrous dysplasia; JOF, juvenile ossifying fibroma; JPOF, juvenile psammomatoid ossifying fibroma; OF, ossifying fibroma; POF, psammomatoid ossifying fibroma; R, retrospective; SBC, simple bone cyst; TJOF, trabecular juvenile ossifying fibroma.

In this context, an ABC appears as a secondary change in a preexisting primary fibro-osseous lesion.[11] The minimum bone structure provided by the fibro-osseous lesion predisposes to rapid expansion and formation of the ABC.[7]

The origin of ABCs continues to be a controversial topic, and it appears to be multifactorial.[1-4,29] The most plausible pathogenic mechanism is related to alterations in vascular homeostatic balance within the bone, resulting in an increase in pressure.[1,2] These alterations may be posttraumatic, reactive, related to genetic predisposition, or hormonal variations, since they may be aggravated by pregnancy.[2,3] Ziang et al affirmed that 50%-70% of patients with an ABC relate a history of trauma.[29] In the present case, the patient denied suffering previous trauma.[29]

Findings from clinical and radiographic examinations can raise suspicion about the association of lesions.[2] Nevertheless, diagnosis requires integrated analyses of the clinical, radiographic, surgical, and histologic data.[2,13,26] In the present case, the characteristics of both lesions were revealed by the histopathologic examination. However, the indications of association in the clinical and radiographic examinations, such as the presence of hemorrhagic liquid on aspiration puncture and the radiopaque images in the center of the radiolucent area, were fundamental in establishing the diagnosis.

The treatment of choice for ABCs is surgical resection, but other therapeutic options have been suggested, such as curettage with bone graft placement, arterial embolization, and intralesional injection of sclerosing agents.[2,3,15] The rate of recurrence related for ABCs varies from 8% to 60%; the greater value is associated with incomplete removal.[30] When fibro-osseous lesions are associated with cysts, the rate of recurrence may increase. Suei et al related recurrence in 90% of simple bone cysts that were associated with bone dysplasia.[31]

The concomitant presence of a fibro-osseous lesion and its subtype usually has an impact on the choice of therapy.[32] Depending on the nature and extent of lesions, the treatment modalities for fibro-osseous lesions, in isolation, may vary from mere observation to enucleation or surgical resection, and the lesion with the worst prognosis dictates the choice of treatment.[33] Fibro-osseous lesions with concomitant cystic degeneration require aggressive measures.[32] In the present case, the procedure indicated for the most serious lesion prevailed: that is, complete removal of the ABC.

Conclusion

The present case emphasizes the fact that lesions with rapid and destructive expansion may represent an ABC secondary to a benign fibro-osseous lesion. This possibility must be considered even when there is no clinical history or radiographic evidence of a fibro-osseous lesion. If there is a history of a fibro-osseous lesion, subsequent rapid expansion is highly suggestive of transformation to an ABC, and not malignant degeneration, as is frequently speculated. These considerations may avoid mutilating surgery for a condition that is radiographically aggressive but histologically benign.

Author information

Dr Jacomacci is a resident in oral and maxillofacial surgery; Dr Perdigão is an oral and maxillofacial surgeon; Drs Veltrini, Farah, and Tolentino are adjunct professors; and Drs Iwaki and Iwaki Filho are associate professors, School of Dentistry, State University of Maringá, Brazil.

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