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CASE REPORT

Aneurysmal bone cyst of the mandible affecting the articular condyle: a case report

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Key Clinical Message

Aneurysmal bone cyst (ABC) is a benign osteolytic lesion that is fast-growing, expansile, and locally destructive. The present case is of a young girl with facial asymmetry, which had become accentuated during the previous months. A conservative treatment was performed to reduce morbidity and affectation of the lower dental nerve.

Keywords

Aneurysmal bone cyst, mandible.

Introduction

Aneurysmal bone cyst (ABC) is a benign osteolytic lesion that is fast-growing, expansile, and locally destructive. These lesions consist of vascular spaces filled with blood, separated by trabecular osteoid tissue and osteoclast-type giant cells [1]. They represent 1.5% of nonodontogenic tumors of the jawbones. They develop in the long bones, the mandible being the most commonly affected area (ABC affects only 2% of upper maxillary bones) [2, 3]. Condylar affectation is extremely rare, and the literature contains only a very few cases [4].

Case Story

This is the case of a 10-year-old girl without antecedents of interest, who came to the Oral and Maxillofacial Surgery Unit of *Virgen de las Nieves* University Hospital (Granada, Spain) presenting facial asymmetry, which had become accentuated during the previous months. Examination identified a left-side facial tumoration in the temporomandibular area, with severe joint dysfunction, lateral deviation, and reduced mouth opening. Intraoral examination found the oral mucosa intact and bulging internal and external cortex of the mandibular ramus. The patient did not present significant neurological disorders. Orthopantomography (OPG) (Figs 1 and 2) revealed a multilobulated radiolucent image with badly defined margins; computerized tomography (CT) (Fig. 3) confirmed the existence of an expansile, multipartitioned, osteolytic, intraosseous lesion in the area of the mandibular condyle and ascending ramus, with bulging and thinning cortex, but without affectation of the soft parts. Inside the lesion were multiple lagoons of vascular appearance, which suggested an initial diagnosis of suspected ABC.

On the basis of these findings, the patient was scheduled for surgery under general anesthetic. At the first stage of treatment, initial embolization of the lesion was performed, removal of the lesion via an intraoral

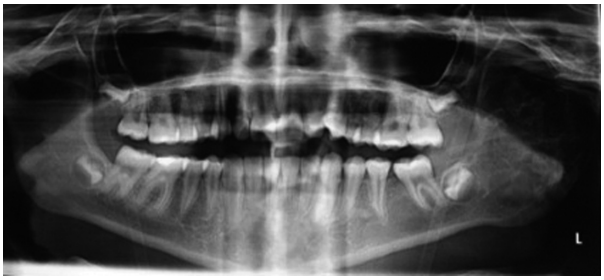

Figure 1. Presurgical orthopantomograph showing radiolucent multilobulated lesion, which covers the mandibular ascending ramus and left condyle.

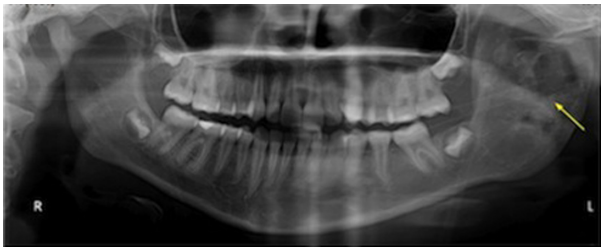

Figure 2. Orthopantomograph of relapsed cystic lesion before the second surgery, which shows the same extension in the area of the ascending ramus and left condyle. Arrow indicates the abnormality.

approach (interpapillary incision in the posterior mandibular sector, extending to the retromolar trigone and the mandibular ramus, subperiosteal stripping, partial osteotomy, and removal of the lesion by means of curettage of the cyst, placing a drainage tube).

Under microscopy, the biopsy corresponded to the peripheral area of the lesion, mostly made up of cortical bone as well as fibrous tissue of an inflammatory appearance, with reactive trabecular bone formation, some of which had a bluish appearance. The area that we interpreted as a central area, although it did not present abundant cystic vascular structures, did present fibrous walls with abundant osteoclast-type multinucleated giant cells. No necrosis, cell atypia, or abnormal mitosis was observed, which would otherwise indicate the malignancy of the material examined. All observations were compatible with an ABC.

One year later, it was found that the lesion persisted, which led to a second surgery to perform further mandibular curettage to achieve complete removal of the lesion and so remission of the disease. The patient presented good postoperative evolution; no local or general complications developed. This conservative treatment was chosen rather than a more radical surgical extirpation due to the patient's young age. This is very important, because with conservative treatment, there was no associated morbidity, but there could have been affectation of the lower dental nerve. Furthermore, there are all the complications inherent to any surgery of this type: mandibular fracture, bleeding, hematoma, infection, etc., but the most important complication would be nerve affectation, while with radical surgery could occur mainly facial deformity (with important consequences for facial aesthetics), malocclusion, and joint dysfunction.

At clinical and radiological follow-up, the disease continued in remission (Figs 4 and 5) with correct joint function and normal occlusion, and without any signs of temporomandibular dysfunction. Facial symmetry had been restored. In magnetic resonance checkups, the disc was seen to be situated in the anatomical position with no signs of luxation. This positive evolution with adequate joint remodeling endorsed the conservative approach adopted (Fig. 6).

Discussion

Aneurysmal bone cyst was first described in 1893 by Van Arsdale, but the term *aneurysmal bone cyst* was not defined and applied until 1942 by Jaffé and Lichtenstein [5, 6]. The etiopathogeny of ABC is controversial and has been a subject of ongoing research ever since the pathology was first described. It remains unclear whether the lesion is primary or secondary to a preexisting bone lesion [6]. Some authors have described it as a congenital lesion; others have claimed that its origin lies in trauma [7, 8], such as a dental extraction with consequent development of subperiosteal hematoma. Other theories relate ABC to vascular origins arising from arteriovenous malformations, which would provoke an increase in intraosseous venous pressure, expansion, or destruction of the vascular bed and bone resorption [9]. Other authors argue that its origin lies in degeneration of a preexisting lesion such as central giant cell granuloma, fibrous dysplasia, hemangioma, eosinophilic granuloma, ossifying fibroma, or chondroblastoma, among others. Other research supports a common origin of central giant cell granuloma with simple bone cyst.

All the theories share a common vascular etiology and the concept that local bone factors will determine pathogenesis [10]. Recent research has also associated ABC with certain chromosomal disorders (16;17) (q22;p13).

In histology, ABC is considered a pseudocyst due to the absence of epithelial walls. It consists of fibrous connective tissue stroma with blood-filled vascular spaces, osteoclast-type giant cells, and osteoids [8]. Three types of ABC can be differentiated histopathologically, with varying vascular components and clinical behavior [4, 11, 12]. The solid type (some 5% of cases) is characterized by dense stroma, scarce vascular spaces, bone

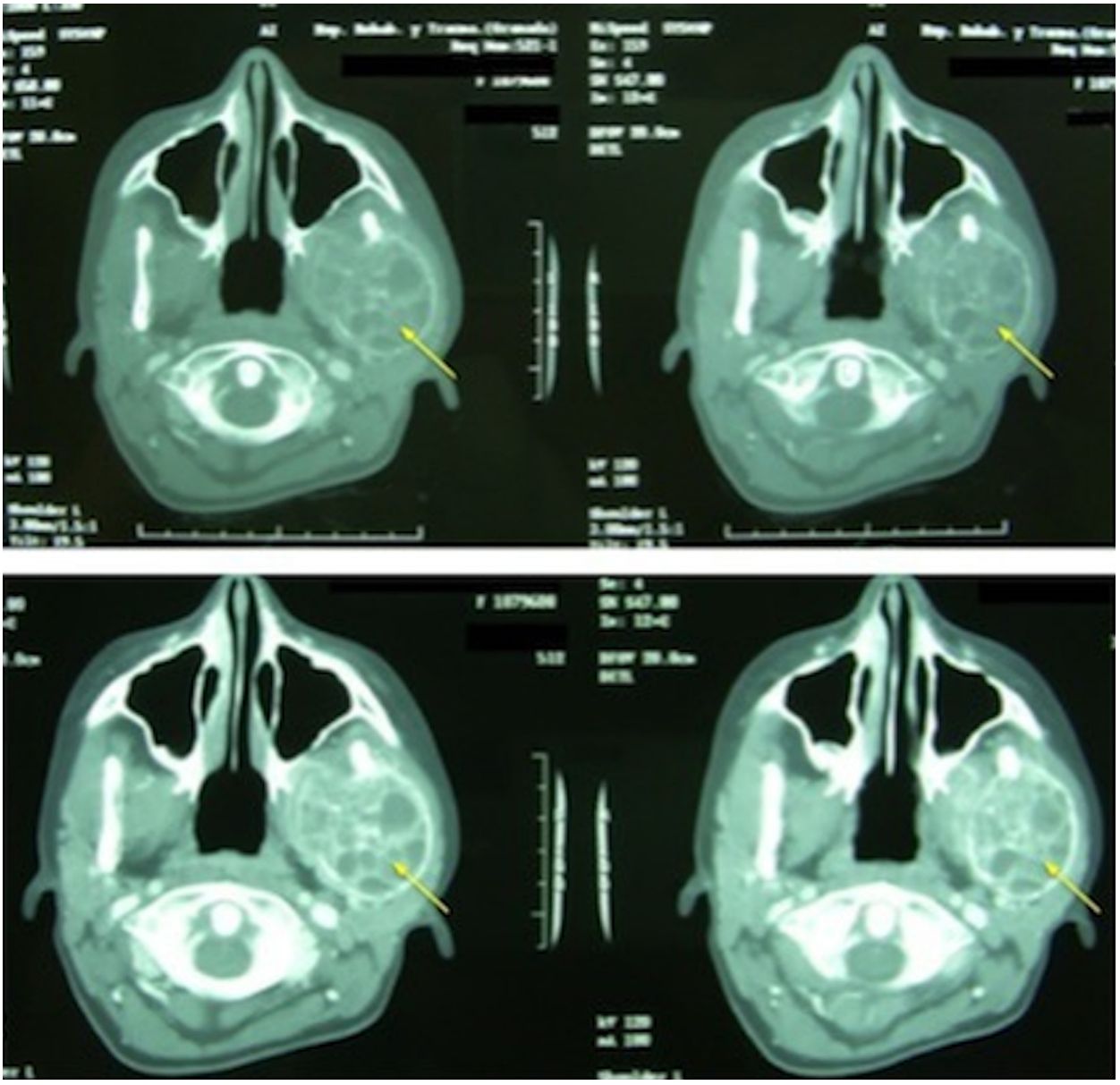

Figure 3. Axial computerized tomograph images showing an intraosseous osteolytic lesion, insufflating and multitabicated, at the left mandibular ascend ramus and condyle, with bulging and thinning cortex, but without soft tissue affectation. Arrow indicates the abnormality.

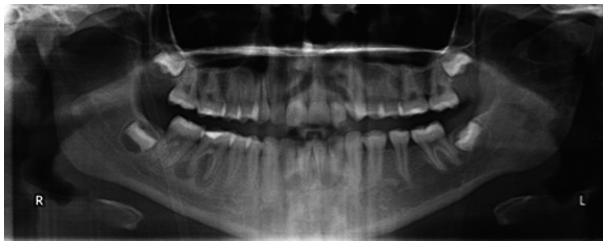

Figure 4. Follow-up orthopantomographs at 3 years.

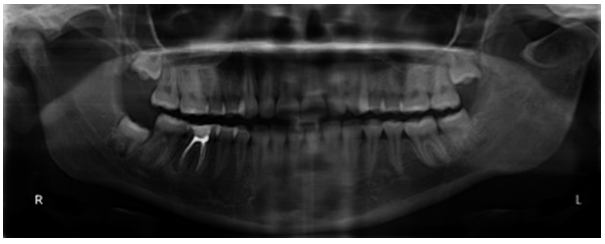

Figure 5. Follow-up orthopantomographs at 7 years, showing the absence of tumor relapse and condylar remodeling.

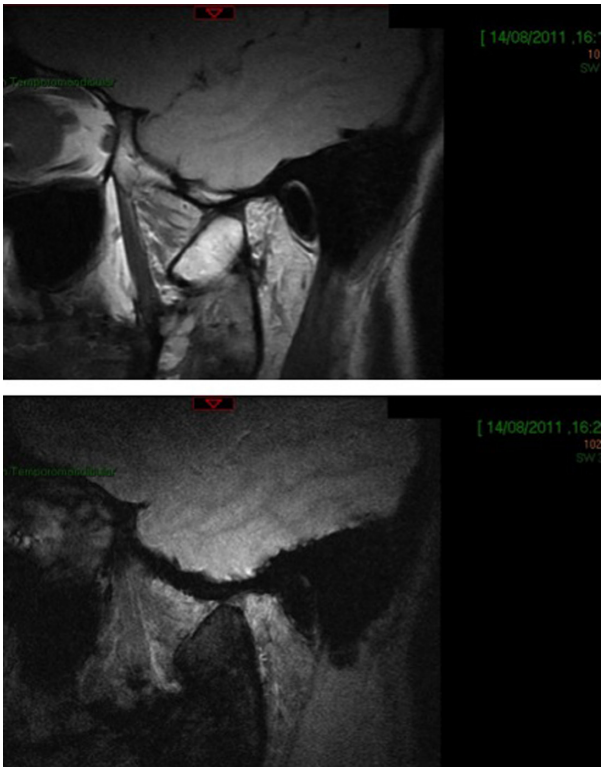

Figure 6. Recent magnetic resonance images show good joint dynamics with the disc situated at its anatomical position.

expansion without perforation, and a low tendency to bleeding during surgery; clinically, it usually presents as an asymptomatic lesion. The vascular type (95% of cases) shows scarce fibrous stroma, numerous dilated, blood-filled vascular spaces, extensive perforation, and bone destruction extending to the soft tissues; during surgery, there will be a strong tendency to bleed. There is also an intermediate ABC variety between the vascular and solid types.

Aneurysmal bone cyst can also be classified according to clinical and radiological manifestations as inactive, active, or aggressive. The clinical signs and symptoms of ABC are diverse and nonspecific. It usually manifests as inflammation of the soft tissues due to expansion of the cortical bone, and secondly the development of facial asymmetry and malocclusion. In the most aggressive cases, great bone destruction takes place, even invading the soft tissues. Other associated symptoms can include pain; paresthesia, mobility, migration, or resorption of the involved teeth; epistaxis; nasal obstruction; proptosis; and diplopia, depending on which area is affected by the lesion.

In the same way as the clinical signs associated with ABC, radiological manifestations are nonspecific and very variable. The typical orthopantomograph image reveals a unicystic radiolucent lesion or multicystic expanse, with cortical bone destruction and an interior trabeculated pattern. The multiloculated pattern endows the X-ray image with a characteristic "honeycomb," "soap bubbles," or "moth-eaten" appearance, which is also characteristic of other lesions such as giant cell central granuloma, myxoma, desmoplastic fibroma, hemangioma, keratocyst, or ameloblastoma. CT is the technique of choice for examining the extent of the lesion and for planning treatment, as it helps to determine the borders of the lesion, while magnetic resonance (MRN) helps to better visualize soft tissue affectation and the presence of liquid content. Angiography is indicated in cases where MRN reveals a hypervascularized lesion.

Under macroscopy, ABC has a spongy appearance, made up of blood-filled cavities separated by fine fibrous partitions. Given the nonspecific clinical and radiological findings, preoperative diagnosis will be confirmed by histological analysis to differentiate ABC from other pathologies that can develop in the maxillofacial region. So both clinical and radiological examinations, together with an incisional biopsy for histological assessment, are essential to diagnosis and treatment planning [13]. In cases with a major vascular component, presurgical angiography and embolization of the lesion will allow better intraoperative hemorrhage management [8, 11, 14].

Given that the clinical signs associated with ABC are nonspecific, there are a variety of possible bone lesions that demand differential diagnosis. Among these are central giant cell granuloma, myxoma, traumatic bone cyst, brown tumor hyperparathyroidism, fibrous dysplasia, ossifying fibroma, desmoplastic fibroma, hemangioma, and ameloblastoma [8, 11].

The treatment of ABC is a controversial subject, and no consensus has been established as to the optimal therapeutic approach; meanwhile, there are a wide variety of equally acceptable options. These include conservative treatment with periodic checkups that aims to achieve spontaneous remission (such as simple curettage), cryotherapy, excision of the lesion, radical resection and reconstruction with bone grafts, therapeutic embolization, or intralesional injections of calcitonin and methylprednisolone [15–19].

The therapeutic option will depend on the size and position of the cystic lesion, the patient's age, the clinical manifestations, and the pathology's extension into the surrounding soft tissues or bone structures (maxillary sinus, nasal cavity) and the particularities of case evolution. In this way, inactive lesions can be treated with a conservative approach or simple curettage, while rapidly progressive active lesions with affectation of the soft tissues and the associated painful symptoms require more aggressive and radical treatment.

The rate of relapse varies between 20% and 30% and it occurs mainly during the first year after surgery. In most cases, this is due to incomplete removal of the lesion, particularly in cases with soft tissue affectation [2, 8, 18]. Some authors have associated a higher rate of relapse with large lesions treated with curettage of the cystic cavity [20, 21].

Conclusions

In the present case, in spite of the lesion's large size, it was decided to adopt a conservative approach in both the surgeries due to the cystic cavity's location, the bone structures involved, and the patient's young age. Bone curettage was performed in both surgeries, obtaining good long-term bone responses with clinical remission of the lesion, facial symmetry correction, and the recovery of joint function, confirmed by MRN checkups. The conservative surgery avoided more radical surgery, which would have involved complete removal of the ramus–condyle complex and its reconstruction, with possible associated morbidity and repercussions for joint function and facial aesthetics.

Condylar affectation resulting from aneurysmal bone cyst of the mandible is very rare, and the existing therapeutic options are very diverse. The choice of treatment is a very controversial subject, and each case must be considered individually, assessing the possible repercussions and morbidity that may develop.

Conflict of Interest

The authors declare that there are no conflict of interests.

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