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CASE REPORT

Unusual radiographic presentation of an aneurysmal bone cyst of the mandible

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Abstract An 11-year-old girl was referred because of a painless firm swelling in the right posterior mandible that had started 2 months previously. A panoramic radiograph showed a nonspecific finding of a tiny discreet shadow following the lower border of the mandible, without any radiographic signs of radiolucency in the affected area or discontinuity of the lower border. However, multislice computed tomography (MSCT) findings were suggestive of an aneurysmal bone cyst, and histopathological findings revealed a diagnosis of aneurysmal bone cyst. Complete surgical excision followed by extensive cortical bone curettage was done, and no recurrence has been observed in the past 5 years. A differential diagnosis list is included, and extended with fibrous dysplasia according to the radiographic findings. To the best of our knowledge, this is the first case of a jaw aneurysmal bone cyst with unusual initial radiographic findings. Furthermore, a ground-glass appearance on MSCT scans suggested fibrous dysplasia. The present case highlights the need for accurate differential diagnosis of the lesion described to obtain the correct diagnosis in a timely manner and plan the appropriate treatment.

Keywords Aneurysmal bone cysts · Jaw diseases · Panoramic radiography · Multislice computed tomography · Fibrous dysplasia

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Introduction

Aneurysmal bone cysts (ABC) arise in 2–12 % of bones within the maxillofacial region, with the mandible being the most frequently affected. The average age of patients presenting with an ABC is 13 years, and 80 % of patients are under 20 years [1]. ABCs are rare, non-neoplastic, and rapidly growing bone lesions that can present with various forms, from slowly progressing to rapidly expanding swellings with pain, deformity, and pressure symptoms [2]. ABCs are characterized by variable clinical and radiographic features [3]. Regarding diagnosis, it appears that only histological findings can allow 100 % certainty. Radiology, magnetic resonance imaging (MRI), and computed tomography (CT) might be suggestive, but are not sufficient to confirm an ABC diagnosis without histology [2, 4]. Three types of ABC based on histopathology have been recognized [5]. A preoperative incisional biopsy is important for the diagnosis prior to surgical treatment [6]. The treatment of ABC is conservative surgical resection [7]. A curettage must be performed. Recurrence of ABC is seen in 21–50 % of patients after simple curettage [8] and in 11–25 % of patients after more radical resection.

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The aim of this report is to present a case of an unusual radiographic appearance of an aneurysmal bone cyst of the lower jaw.

Case report

An 11-year-old female patient was referred to the Department of Oral and Maxillofacial Surgery with the chief complaint of a painless, hard, and tender swelling of the right posterior mandible. Her detailed medical history revealed that the swelling had started 2 months previously, and subsequently increased to the present size over the previous month. Medical records and family history were unremarkable, and the patient had no previous trauma in the swollen region. The absence of symptoms such as fever or suppuration was suggestive of non-odontogenic infection.

Extraoral examination revealed a well-defined, tender, and bony-hard mass at the lower and posterior part of the mandibular body on the vestibular side. The anteroposterior diameter of the lesion was approximately 2 cm, and the superoinferior distance was approximately 3 cm from the inferior border of the mandible. An obvious facial asymmetry was present without any pathology of the overlying skin. There were no signs of inflammation, bleeding, or pus discharge. Paresthesia in the area innervated by the right lower alveolar nerve was not present.

Intraoral examination revealed a tender, well-defined, and bony-hard expansion in the posterior vestibular area of the right side of the lower jaw. The mucosal tissue was intact, with no sinus opening or discharge. There were no carious, discolored, or fractured teeth. The teeth in the involved area gave normal responses to electric and thermal pulp vitality tests.

A panoramic radiograph of the patient's jaw was nonspecific (Fig. 1), revealing a relatively ill-defined radiolucency of approximately 4.2 × 2.8 cm in the body of the mandible, placed under and between the canine, premolar, and molar roots. There were no radiographic signs of discontinuity of the lower border. No lateral or apical root resorption, tipping, or displacement of the teeth was noted.

Taking into consideration all of the above findings, which were clinically and radiographically nonspecific for odontogenic infection, cyst, and tumor lesions, a fine-needle aspiration biopsy was performed. The findings were nonspecific. Multislice computed tomography (MSCT) scans (Fig. 2a, b) and "bone window" 3D reconstruction using caudocranial projection (Fig. 3) showed a well-defined expansile subperiosteal pathological lesion along the inferior border of the right mandible, with a length of

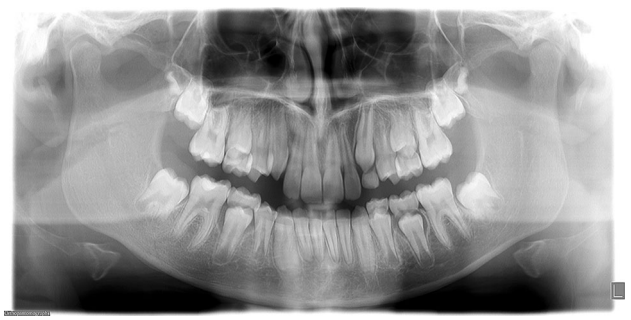

Fig. 1 Nonspecific initial digital orthopantomogram. A panoramic radiograph revealed a relatively ill-defined radiolucency of approximately 4.2 × 2.8 cm in the body of the mandible, placed under and between the canine, premolar, and molar roots, with continuity of the lamina dura of all roots. No lateral or apical root resorption, tipping, or displacement of the teeth was noted

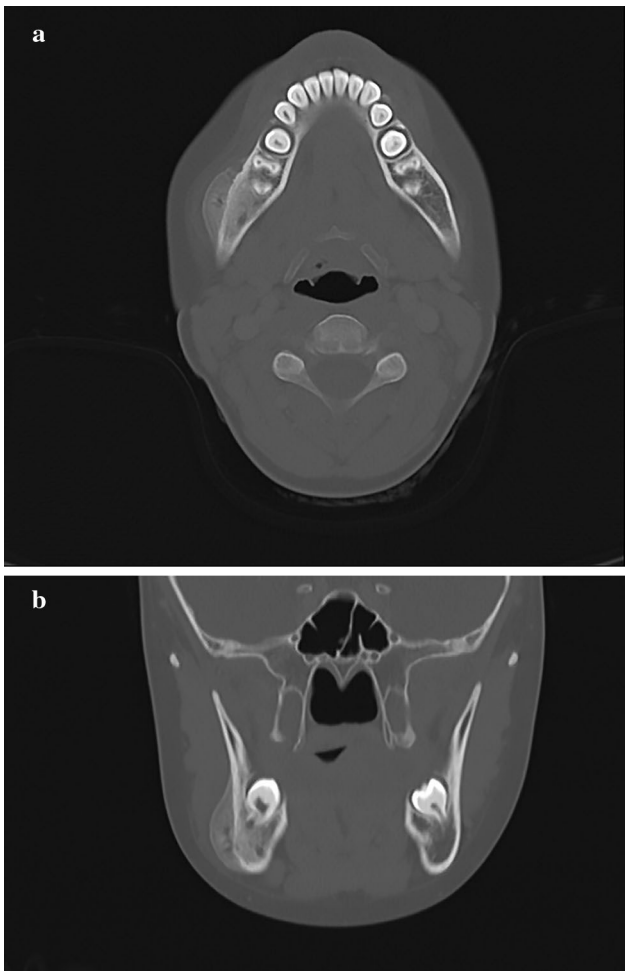

Fig. 2 MSCT scans. **a** An axial section showed a well-defined expansile intraosseous lesion with a ground-glass appearance, which was partially pressed and extended into the perimandibular cheek soft tissue, but without signs of soft tissue infiltration. The intraosseous part of the lesion had relatively ill-defined edges and showed increased cortical sclerosis, without any certain signs of cortical destruction. **b** A coronal section showed that the extraosseous edges of the lesion were radiologically well-defined

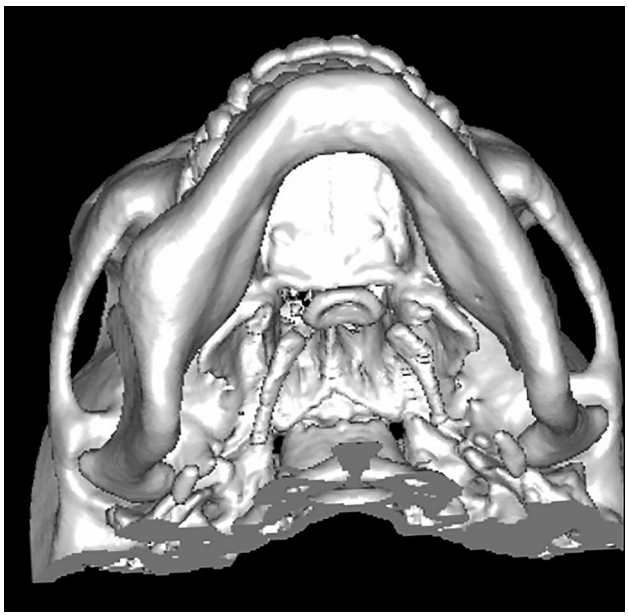

Fig. 3 Volume-rendering MSCT (caudocranial projection) showed an expansile lesion in the posterior part of the mandibular body (towards the angulus of the mandible) deforming the mandibular contours

2.6 cm, height of 3.2 cm, and thickness of 0.9 cm. The lesion was described as a dome-shaped protrusion with a ground-glass appearance, which was partially pressed and extended into the perimandibular cheek soft tissue, but without any signs of soft tissue infiltration. The intraosseous part of the lesion had relatively ill-defined edges and showed increased cortical sclerosis, without any certain signs of cortical destruction. The extraosseous edges of the lesion were radiologically well-defined. The ground-glass appearance suggested fibrous dysplasia disease. Minimal changes without a break in the continuity of the cortical and spongious bone of the mandible were observed. There were no pathologically increased lymph nodes.

Considering the clinical presentation, radiographic features, and nonspecific findings of the aspiration biopsy, an excisional surgical treatment was performed. Under general anesthesia, the lesion was exposed through an extraoral submandibular approach, and a complete surgical excision followed by an extensive cortical bone curettage was performed. The excised specimen was sent for histopathological examination. Prophylactic antibiotics and analgesics were administered for the next 5 days.

Histopathological analysis revealed trabecular bone fragments with osteoblastic rimming within a vascularized loose fibrous tissue, covered by erythrocytes and fibrin, consistent with an aneurysmal bone cyst wall (Fig. 4). In addition, numerous small and large vascular spaces lined by endothelial cells and abundant pools of red blood cells were seen. Therefore, the diagnosis of an aneurysmal bone cyst was confirmed by histopathological evaluation.

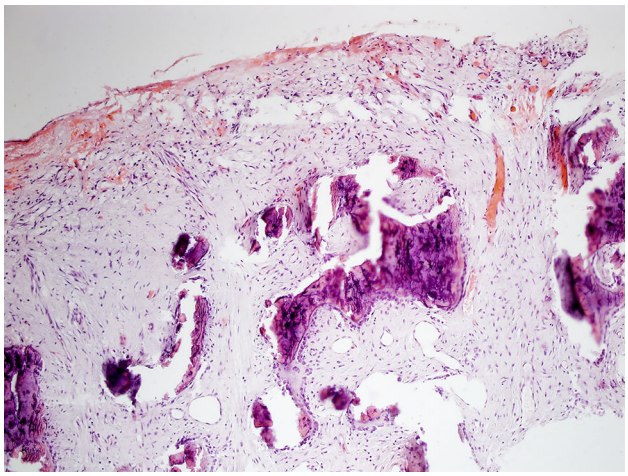

Fig. 4 Histopathological examination of the specimen revealed trabecular bone fragments with osteoblastic rimming within a vascularized loose fibrous tissue, covered by erythrocytes and fibrin, consistent with an aneurysmal bone cyst wall. Numerous vascular spaces lined by endothelial cells and abundant pools of red blood cells were observed

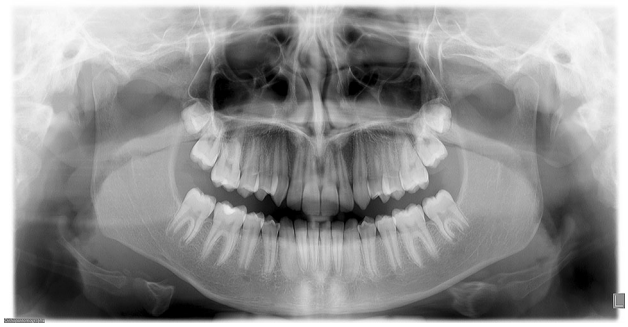

Fig. 5 A panoramic radiograph on follow-up taken 2 years postoperatively revealed normal findings, without any evidence of relapse

A panoramic radiograph (Fig. 5) and latero-lateral teleradiograph (Fig. 6) on follow-up at 2 years postoperatively revealed normal findings, without any evidence of relapse. The patient has been in regular follow-ups for the last 5 years with no evidence of recurrence.

Discussion

Motamedi et al. [9] reported that ABCs had variable presentation, disease course, and histopathologic type, with no sex predilection. ABCs were significantly more common in childhood and adolescence, as well as in the mandible and posterior areas of the jaws. Of the cases reviewed, 90 % were treated by excision and curettage. ABCs had a

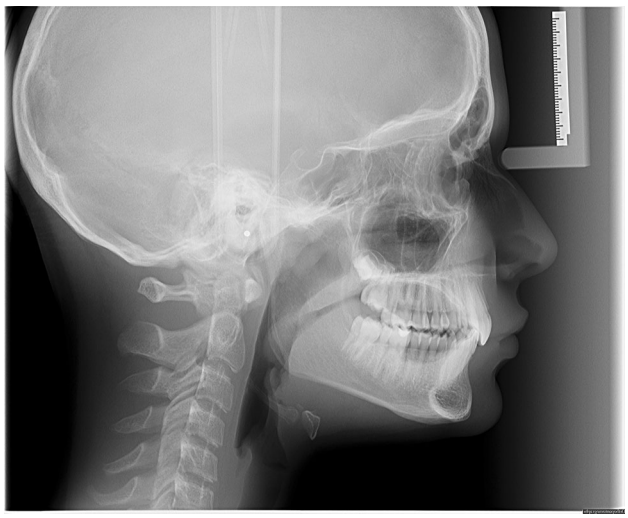

Fig. 6 A lateral teleradiograph taken 2 years postoperatively revealed normal findings, without any evidence of relapse

relatively low recurrence rate (<10 %), precluding the need to perform aggressive surgery primarily.

It has been suggested that a trauma might precede the development of ABC. However, our patient and her parents denied any mandibular trauma in the past. Fortunately, no recurrence was observed in the patient during the following 5 years after surgery. Recurrences mostly happen within the first 2 years after surgery. Zadik et al. [10] reported temporomandibular dysfunction in 50 % of patients with ABC, which was not the case with our patient.

Lately, a case of a large ABC in the ascending mandibular ramus was described by Neuschl et al. [11], wherein a free fibula flap was used to reconstruct the affected area.

Lee et al. [8] highlighted the need for a differential diagnosis, both radiologically and histopathologically, because ABCs can easily be interpreted as giant cell tumors or osteoblastomas, and, on occasion, be mistaken for osteogenic malignancies. Parashari et al. [12] described four ABC cases presenting with different imaging modalities, which were later confirmed by histopathology. Nevertheless, none of the indicated cases showed non-specific initial radiographic features, as observed in the present case. However, in the present case, the radiographic findings were suggestive of fibrous dysplasia. MSCT scans and axial and coronal "bone window" 3D reconstructions showed a well-defined expansile intraosseous lesion with a ground-glass appearance, which suggested fibrous dysplasia disease. To the best of our knowledge, this is the first reported case of ABC of the mandible whose radiological findings potentially indicated fibrous dysplasia, and our recommendation is to include fibrous dysplasia in the differential diagnosis list for ABC in the future.

Henriques et al. [13] evaluated nine patients with ABC. A painful swelling was the most common clinical finding (4/9), which is completely contrary to the present case. Radiologically, the lesions frequently presented as multilocular (5/9), well-defined (4/9), and bone expansion and perforation (2/9). The present case is unique, because the patient had normal orthopantomogram findings, i.e. there was no unilocular or multilocular expansion. Pathological analysis revealed that two cases were associated with a central ossifying fibroma and one case with a central giant cell lesion. Histomorphology showed predominance of the solid type (5/9) and sinusoidal pseudocystic spaces (4/9). Giant cells, osteoid material, calcified material, blood vessels, and hemosiderin deposits were observed in 6/9, 7/9, 8/9, 9/9, and 7/9, respectively. The patients with ABCs presented clinical and radiographic features that often posed a diagnostic dilemma. Knowledge about the most common characteristics of ABCs may contribute to the establishment of a more accurate diagnosis. Sun et al. [14] reported that most jaw ABCs are secondary in nature and frequently associated with ossifying fibroma. However, this is contrary to our findings, as there was no such association.

Common differential diagnoses might be dentigerous cyst, radicular cyst, simple maxillary cyst, fibrous dysplasia, giant cell reparative granuloma, ameloblastoma, cementifying fibroma, odontogenic keratocyst, odontogenic myxoma, solitary bone cyst, and telangiectatic osteosarcoma [2].

In a study on 12 patients, Srinivasan et al. [15] reported that the inferior alveolar canal on the normal side was visualized as a hyperintense structure in relation to the hypointense bone on curved multiplanar reformatted (MPR) images reconstructed from volume interpolated breath-hold examination (VIBE) images in all 12 patients. In nine patients, the inferior alveolar canal was equally well visualized on panoramic CT and curved MPR VIBE images. In two patients, the inferior alveolar canal was better visualized on curved MPR VIBE images, while in one patient, the course of the mandibular canal was better seen on panoramic CT images. MR reconstructions with VIBE sequencing as source images provide images that are comparable to CT reconstructed images for evaluation of the mandibular canal. Three-dimensional (3-D) VIBE sequencing can be added to the MRI protocol to visualize the inferior alveolar neurovascular bundle. 3-D VIBE sequencing increases the diagnostic capabilities of MRI when used to image mandibular cysts and tumors.

To the best of our knowledge, this is the first case of a jaw aneurysmal bone cyst with unusual initial radiographic findings, and therefore highlights the need for accurate differential diagnosis of the lesion described to obtain the correct diagnosis in a timely manner and complete appropriate treatment planning.

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Compliance with ethical standards

Conflict of interest Dragana Gabrić, Spomenka Manojlović, Dijana Zadravec, Vanja Vučićević Boras, and Mišo Virag declare that they have no conflict of interest.

Human rights statement and informed consent All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1964 and later versions. Informed consent was obtained from the patient's parents to publish the case as a clinical report.

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