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Case Report

Aneurysmal Bone Cyst of Mandibular Condylar Neck and its Management - A Fortuitous Appearance in Pediatric Population

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Abstract

The aneurysmal bone cyst (ABC) is a benign, blood-filled bony lesion that commonly affects the vertebral column and long bones of the body. ABCs can appear in the head and neck on rare occasions. ABCs have the ability to grow quickly and weaken the associated bone, resulting in pathological fractures as an early symptom. ABCs are usually asymptomatic, but they can cause local pain and swelling. Only 2-3% of ABCs occur in the head and neck region, with the mandible and maxilla being the most common sites of occurrence.

Aneurysmal bone cysts can be surgically or non-surgically treated. Surgical intervention, regardless of where the ABCs are located in the body is the mainstay of treatment. En-bloc resection is the gold standard of treatment for ABCs because it has the lowest risk of recurrence and also because majority of ABCs are found elsewhere other than head and neck. More delicate surgical techniques, such as curettage with bone grafting, are preferred due to the delicate nervous and vascular structures found in the head and neck region, as well as aesthetic considerations.

The present paper highlights the screening, examination and management of two cases of ABC that were accidentally diagnosed during routine dental checkup.

Keywords: Aneurysmal Bone Cyst (ABC); Decompression; Cysts of Mandibular Condyle

Abbreviation

ABC: Aneurysmal Bone Cyst; JABC: Juvenile Aneurysmal Bone Cyst; KCOT: Keratocystic Odontogenic Tumor

Introduction

The aneurysmal bone cyst of the jaw is a type of pseudocyst. It is a non-neoplastic bone lesion characterized by the replacement of fibro-osseous tissue with blood-filled sinusoidal or cavernous spaces. The lesion is still a relatively uncommon finding in the facial bones, etiology and pathogenesis are unknown and they are most commonly found in the metaphysis of the long bones and the vertebral column [1]. Jaffe and Lichtenstein coined the term aneurysmal bone cyst in 1942 to describe two cases of erosive,

Expansile, blood-filled cystic lesions in an 18-year-old boy's vertebra and a 17-year-old boy's pubic symphysis [2]. Around 2-3% of these lesions are found in the head and neck region, with mandible being the most common location [1]. ABC was thought to be a pseudocyst of hemodynamic or reaction origin, in other words, "primary ABC" or "secondary" to other diseases. Any traumatic cause that results in intra medullary or sub periosteal haemorrhage, followed by an incorrect repair process, or other hemodynamic states, can result in an enlarging vascular bed and bone erosion. The term "secondary aneurysmal bone cyst" refers to a hemorrhagic reaction in a pre-existing bone lesion. The initial lesion may be completely replaced by a secondary ABC or it may remain a part of it [4]. This uncertainty makes distinguishing between a pure ABC and an affiliated ABC more difficult, even though histopathology is critical in the final diagnosis [3]. The majority of ABC cases are asymptomatic, but swelling and local pain may occur [4]. Surgery, such as complete excision, curettage, and bone grafting, is currently the treatment of choice for ABCs [5]. The aneurysmal bone cyst's also have characteristics, such as sudden growth, bicortical expansion, cortical destruction, osteoid formation, and tumour - like appearance, can easily be confused with malignancy.

There is very less literature on ABC in pediatric head and neck region indicating that these clinical entities are underserved. ABC in children occurs very rarely in the jaws, representing about 1.5% of all non-odontogenic and non-epithelial cysts of the jaws. ABC represent only 0.5% of all the jaw cysts. The average age of occurrence is 13 years and 80% of patients are less than 20 years old with no gender predilection. An aneurysmal bone cyst of the condyle is even more unusual, with only 6 cases reported in the literature [6].

Case Presentation

We report the clinical manifestations and management of ABC of mandibular condylar neck in two pediatric patients with similar radiological features, and also the review of literature to further discuss about diagnostic and treatment challenges.

Case I

A 14 year old healthy female visited a dental clinic with chief complaint of prominent lower jaw. OPG and Cephalograms were advised for radiological assessment. Upon radiographic examination a well-defined oval shaped radiolucent lesion was detected in the left condylar neck region by chance. Mouth opening was adequate with no history of pain, discomfort or trauma in that region. A Cone beam computed tomography (CBCT) was obtained, which presented a unilocular well defined radiolucent lesion involving neck of the left mandibular condyle measuring approximately about 18.8 X 8.4 mm with circumferential expansion and thinning of cortical plates suggestive of cystic/benign lesion (Figure 1).

Figure 1: Pre-operative radiograph showing well defined radiolucent lesion in the left mandibular condyle neck region.

Figure 2: Intraoperative photograph showing empty cavity with a thin epithelial lining.

Figure 3: Post-operative radiograph showing adequate bone formation and regression of the cyst after one year.

Case II

A 14-year-old male reported to our center with a history of mild pain and swelling in front of his left ear. Overlying soft tissue was normal and mouth opening was adequate without deviation except tenderness over left pre auricular region. Patient gave no history of trauma in that region. Cone beam computed tomography (CBCT) was obtained which revealed a unilocular well defined radiolucent lesion involving neck of the left mandibular condyle measuring approximately 31.5X16.5mm with similar way of cortical plates thinning and expansion suggestive of a cystic/benign neoplastic lesion.

Figure 4: Pre-operative radiograph showing well defined radiolucent lesion in the left mandibular condyle.

Figure 5: Intraoperative photograph showing empty cavity with a thin epithelial lining.

Figure 6: Post-operative radiograph showing adequate bone formation and regression of the cyst after one year.

Based on radiographic findings, a keratocystic odontogenic tumor (KCOT)/central giant cell granuloma/cystic changes in a fibro-osseous lesion and ameloblastoma were considered in the differential diagnosis of above mentioned cases (Figure 4).

Clinical workup

Incisional biopsy or needle aspiration was not preferred as the lesions in both the cases were not under direct surgical access, keeping in view the possibility of vascular component within the lesion. Hence it was decided to explore the lesion under general anesthesia with a working plan of enucleation in case of cystic

lesion or resection of the affected condyle and reconstruction of ramus- condylar unit with a costochondral graft in case of benign tumour avoiding second surgical procedure.

Surgical workup

Both the patients were proposed surgical procedure under general anesthesia. Pathological site was exposed via Risdon's incision approach. The outer cortex was thin with visible expansion and a pinkish hue indicating a cystic lesion. Therefore, needle aspiration with 18 gauge needle was performed. There was no positive aspirate of blood, cystic fluid or pus except for a drop of blood coated to the hub of the syringe. Later buccal cortical window/outer cortical de-roofing over neck of the condyle was done to enter the cavity, where we found scanty blood clot and a thin friable lining, which was carefully curetted and sent for histopathology. Since it was an empty cystic cavity in both the cases suggestive of either ABC or traumatic bone cyst was packed with absorbable gel foam and the surgical site was closed in layers instead of resection and reconstruction. Final Histopathological reports suggested ABC in both the cases. Patient was maintained on liquid/soft diet for 2 months to facilitate uneventful healing (Figure 2 and 4).

Treatment outcome

Both the patients were kept under periodic follow-up. Extra oral wound healing was uneventful. Radiographs were taken at an interval of 3 months that showed reduction in the size of the cystic cavity with no further expansion. By the end of 1 year a CBCT was obtained which presented satisfactory bone formation within the cystic cavity providing adequate shape and strength to the condyle of mandible (Figure 3 and 6).

Discussion

Aneurysmal bone cyst is definitely a non- neoplastic lesion, and no evidence exists to suggest that trauma has any role other than to signal a preexisting lesion [7].

The cause of aneurysmal bone cyst remains unclear, although there are several theories on it. Lichtenstein - proposed that the lesion originates in a circulatory disturbance, which leads to a dilated and congested vascular bed. This engorged vascular bed then causes resorption of spongy bone, erosion of cortical bone, and expansion of the lesion, which is why it is described as "aneurysmal." Biesecker., *et al*. proposed that formation of aneurysmal bone cyst is secondary to a primary benign bone lesion. The primary lesion prompts arterio-venous malformation, thereby creating a secondary reactive bone lesion (known as aneurysmal bone cyst), which is supported by a series of 66 cases in which a primary bone lesion. Bernier and Bhaskar '' proposed that aneurysmal bone cyst begins as a focus of intra medullary haemorrhage and that the lesion represents an unusual reparative response to haemorrhage. If a connection is maintained between the hematoma and the damaged blood vessel, aneurysmal bone cyst results. If the connection is obliterated, giant cell reparative granuloma results [8].

Juvenile ABCs can affect people of any age, however the first two decades have higher occurrence. Typically there is no gender predominance, and the susceptibility to damage is more in mandible having more than 90% of JABCs occurring in the posterior aspect. JABC can have a wide range of clinical characteristics, from asymptomatic lesions that infrequently appear as radiolucencies on standard radiography to occasionally expanding and destructive patterns. According to the available data, a painless swelling is the primary sign of JABCs [7]. Pain and/or edema are the major symptoms [9]. Rapid growth has frequently been documented and less frequently, lesions may throb or feel as though they are under pressure, and occasionally they may even pulsate [9]. Headache, diplopia, blurred vision, proptosis, loosening of the teeth, hearing loss and nasal obstruction have all been reported as additional symptoms.

The lesion may resemble a malignant tumour due to its rapid and destructive growth which is accompanied by significant osteolysis and cortical damage [9]. Initial lesions are tiny and are less likely to expand the bone but could have a concerning permeative pattern. The lesion is medullary-based, eccentric or centric, always exhibits osteolysis, resulting in expansion of the host bone, and almost always has a pattern of geographic distribution.

The expansion and thinning of the cortex as well as the enlarged, modified "blown-out" or "ballooned" body contour of the host bone are frequent effects of ABC growth. Sharp, well-defined, trabeculated margins without signs of periosteal response are also typical of ABC [10]. Both the cases in the present paper represented similar pattern as mentioned above.

Radiologically, the lesions often appear as radiolucencies with expansion and thinning of the cortical plates in most of the cases (87%), but can also be radiopaque (2%) and have a mixed appearance (11%) [9]. These lesions do not have a recognizable radiographic appearance, and histology is used to make the diagnosis. It must be taken into account that osteoid tissue is a normal part of the fibrous septa and that there may be many mitotic figures in ABC even if anaplasia and atypical mitoses are absent. Additionally, a true giant cell tumour may contain a secondary ABC component [9].

Typical ABC have a sharp, well-defined and trabeculated margins with no evidence of periosteal reaction. Three patterns of involvement have been reported: eccentric (most common in large tubular bones), parosteal (the least common), and central. Also, ABC may exist as an independent (primary lesion) or may be associated with another condition (secondary ABC).

After examination, a differential diagnosis can be given as giant cell tumour, telangiectatic osteosarcoma, unicameral bone cyst, nonossifying fibroma, ossifying fibroma and cementifying fibroma, enchondroma, fibrous dysplasia, myxoma, bone fracture, fibroblastic osteosarcoma, and reparative giant cell granuloma of hyperparathyroidism [10,14,15].

How ABCs should be addressed depends on the patient's age, location, size and extent of the lesion, and any lingering concomitant conditions. A biopsy is the most important step for diagnosis prior to treatment. Since both the cases reported with lytic Expansile lesions located in the condyle of the mandible, the safe option was to perform aspiration/incisional biopsy under general anesthesia. However this would necessitate a second procedure under general anesthesia for definitive management. Since the diagnosis was not confirmed and there was a possibility of a vascular lesion, it was decided for direct exploration of the lesion and on table decision of specific treatment.

Curettage is a simple procedure to cure dormant lesions. Although sclerosing agent injections or selective arterial embolization may greatly help with treatment, they are hardly ever documented in JABCs. Since ABCs frequently afflict children, cautious curettage is recommended as the primary treatment [11]. If the lesion progresses, it becomes painful, or distrusts bone structures then resection should be taken into consideration. Marginal resection may be done in tiny lesions with low morbidity and recurrence risk [11]. Although there is a real risk of recurrence, it can be controlled with local adjuvant therapy like cryotherapy and sclerosing agent injection. In extreme cases, the cyst position and size may make them extremely risky for surgical treatment, making resection impossible and curettage difficult due to the risk of haemorrhage. As a type of treatment, the defect is fixed through grafting or complete surgical excision. Curettage recurrence rates range from 20 to 70% [9], while resected lesions experience recurrence rates of 11 to 25% [11]. Recurrences have been common, probably due to the difficulties in entirely eliminating very large lesions. When surgery is technically difficult or not recommended for people at high risk, percutaneous sclerotherapy is a significant option. Despite the fact that radiation can be used, a radiation sarcoma may occur [9,12].

In both the cases reported in this article, on exploration under GA it was an empty cystic space which was suggestive of ABC and Considering various treatment options, Curettage was done and patient was kept under close observation for a year. This follow up period showed satisfactory bone formation and no recurrence was noted. These two case studies suggest that early detection and curettage can definitively eradicate this aggressive bone lesion.

Conclusion

Aneurysmal bone cysts, which can be surgically or non-surgically treated, require early detection and management. The mainstay of treatment is surgical intervention, regardless of where the ABCs are located in the body. Alteration of the dynamics of the lesion by de-roofing or creating a window and gentle curettage facilitate uneventful healing. Such lesion in the condylar region warrants extra oral approach under general anesthesia. Therefore, we recommend surgeon to be prepared to perform definitive treatment immediately once it is explored under general anesthesia to avoid second surgical procedure.

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