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Case Series

Condylar Aneurysmal Bone Cyst in Paediatric Patients: Series of 3 Cases with Literature Review

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Abstract

Aneurysmal bone cysts (ABCs) involving condyle is an unusual clinical presentation with only 11 cases below 12 years of age reported till date in English literature. We present a series of three paediatric patients with condylar ABCs and focus on diagnostic and therapeutic approaches in children based on our own experience and a critical review of the literature. In our cases of ABC condyle, resection was followed by immediate reconstruction of ramus condyle unit with costochondral graft as a primary choice. Minimum follow-up period was of one year was done to confirm a successful functional and aesthetic outcome of the treatment.

Keywords: *Aneurysmal Bone Cyst; Paediatric; Mandibular Condyle; Costo-Chondral Graft*

Introduction

The World Health Organisation (WHO) classifies aneurysmal bone cysts (ABCs) as non-neoplastic, benign and locally expanding osteolytic tumour-like lesions. These lesions consist of blood-filled spaces separated by trabeculae of osteoid tissue with osteoclast giant cells [1]. The major locations of aneurysmal bone cysts are metaphysis of long bones like the femur (more than 50%) and the tibia (12 - 30%) or the spine [2]. Only 1 - 3% of the lesions are located in the craniofacial complex [3]. Among these, condylar involvement is extremely rare, and the literature reports only few cases. Aneurysmal bone cysts show varied clinical symptoms, and literature describes indolent and asymptomatic presentations as well as painful, rapidly growing and destructive lesions [4-7]. We report a case series of condylar -ABC in children below 12 years of age with a specific emphasis on the diagnostic and therapeutic challenges in this paediatric population.

Case Reports

Case 1

A 11-year-old girl was referred to our maxillofacial unit with a seven-month history of gradually increasing painless right preauricular swelling (Figure 1). Physical examination revealed a swelling of approximately 3 cm in diameter over the right preauricular region which was firm to hard and non-tender on palpation. A mouth opening of 29 mm was noted. An aspiration biopsy with cytologic evaluation from the swelling yielded blood mix fluid smear revealing degenerated lymphocytes in a haemorrhagic background.

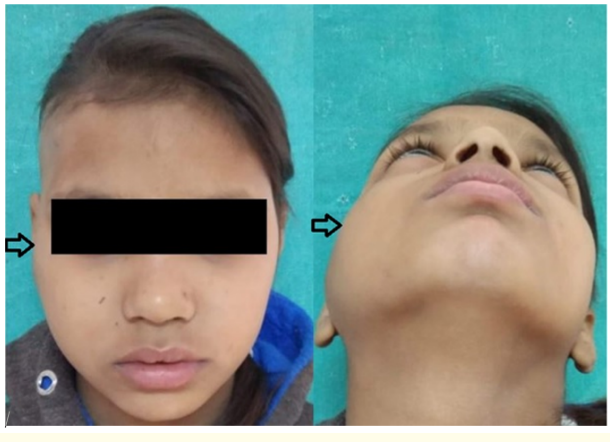

Figure 1

A contrast enhanced computed tomogram (CECT) of the mandible with Angiography showed a well-defined solitary multilocular expansile lytic lesion involving the right mandibular condyle and ramus measuring 3.3 (Anteroposterior) X 3.2 (Transverse) X 2.5 (Craniocaudal) cm in size (Figure 2). The lesion resulted in focal

areas of cortical breach posteriorly in condylar process. Magnetic Resonance Imaging (MRI) revealed heterogenous fluid filled internal areas showing T2 w hyperintensities and T1 hypo intensity along with septations. The lesion produced a mass effect displacing the pterygoids medially and masseter laterally.

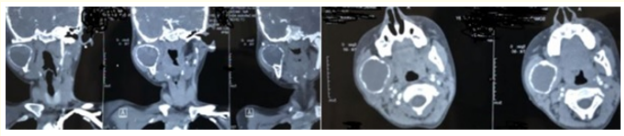

Figure 2

Case 2

An 8-year-old girl presented with history of swelling in front of the left ear, which had progressively aggravated over the past 8 months. Clinical examination revealed a swelling of roughly 4 cm diameter in the left preauricular region which was non tender and relatively firm on palpation with a smooth overlying skin surface. Intraorally, the swelling was associated with expansion of both lingual and buccal cortices in the ramus. The patient's interincisal opening was 30 mm. The range of motion in the left temporomandibular joint (TMJ) was decreased but painless, and there was a slight mandibular laterognathism on opening towards affected site. Aspiration cytology of the tissue obtained from the lesion showed numerous RBCs and scanty inflammatory cells.

CECT of the mandible with Angiography revealed bony expansile lytic lesion measuring 4.26 (Anteroposterior) X 3.61 (Transverse) X 4 (Craniocaudal) cm in size involving the left mandible, extending superiorly till the coronoid and condylar process. Inferiorly the lesion extended up to the mandibular arch with an impacted 37. The lesion was closely abutting the left maxillary artery along the postero-medial wall of condyle with cortical thinning and perforation at various areas.

Case 3

A 10-year-old boy reported with left pre-auricular swelling of 5-month duration associated with dull pain. Extraoral examination revealed a hard 5-cm tender swelling over the left preauricular region. The mouth opening was 32 mm without clicking or crepitation, but the mandible deviated to the left on opening. There was no sensory impairment in the preauricular region. Intraorally there was a diffuse compressible swelling involving the left hemimandible extending up to the retromolar region. Aspiration cytology revealed hemosiderin laden macrophages in a background of RBCs.

CECT mandible with Angiography revealed a well-defined expansile lytic lesion measuring 4.5 (Anteroposterior) X 3.7 (Transverse) X 6 (Craniocaudal) cm in size arising from the crown of the left 3rd molar tooth involving the left hemimandible, extending superiorly up to the condylar process. Few areas of cortical breach were noted on the lingual side of the lesion.

None of the three cases had any history of trauma or any other relevant familial, medical or dental history.

Diagnosis

The clinical and radiologic features in these patients presented a spectrum of possible diagnoses. Consequently, our differential diagnosis included Osteolytic lesions of the ramus condyle unit (RCU) such as primary odontogenic/nonodontogenic cysts and tumours, fibro-osseous lesions and vascular malformations. To confirm the diagnosis, a resection of the involved RCU of mandible was planned for all 3 cases as an excisional biopsy, followed by immediate reconstruction with costochondral graft.

Surgical protocol

In all cases, the surgery was performed under general anaesthesia. The lesion on the affected side was exposed via an extended submandibular approach (Figure 3). Complete resection of the ascending ramus including the coronoid and condylar process was performed leaving the condylar disc intact. Maxillomandibular fixation (MMF) was done to secure the occlusion intraoperatively. A costochondral graft from the fifth rib was harvested, its length was determined intraoperatively by the size of the resected specimen. Graft fixation at the recipient site was done using a low-profile locking titanium reconstruction plate (DePuy Synthes®, Paoli USA), such that the chondral end of the graft was abutting against the articular disk. All specimens were fixed in 10% formalin and embedded in paraffin with gross specimen showing expansion seen of cortices and perforation at various sites (Figure 3).

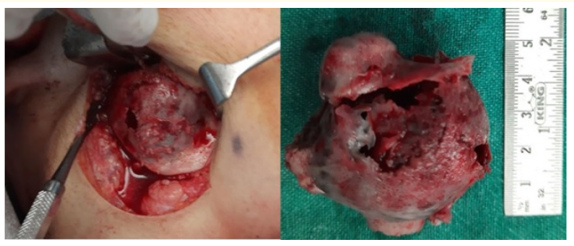

Figure 3

Follow-up

Post-operatively, maxillomandibular fixation was maintained for a period of two weeks. After its release, children were motivated for active mobilization and mouth opening exercises. All patients were followed up at regular intervals for a minimum period of 12 months. At all stages of clinical and radiological follow-up, these patients showed no sign of recurrence (Figure 4). with a favourable joint function and normal occlusion, without any signs of temporomandibular dysfunction. Facial symmetry had also been restored.

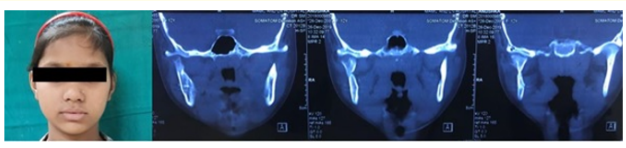

Figure 4

On histological examination, the tumour was found to be composed of fibrous tissue, blood, osteoclast-type giant cells and reactive woven bone (Figure 5). Based on these findings, a diagnosis of Aneurysmal Bone Cyst of the condyle was established.

At the end of the follow-up period at 12 months, a non-contrast CT was done for all 3 cases which revealed excellent graft uptake and a favourable joint morphology at the affected site.

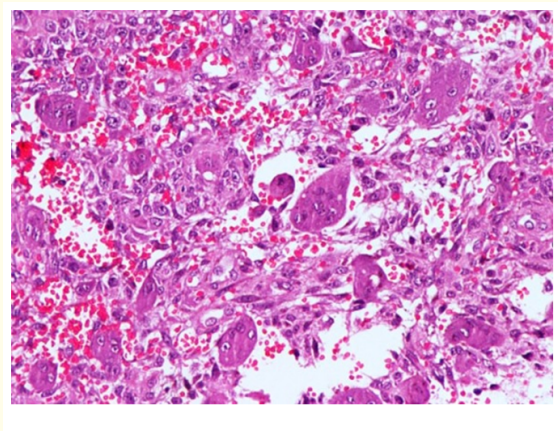

Figure 5

Discussion

Aneurysmal bone cysts were firstly described in 1893 as a "homerus ossifying haematoma" [8]. Later the term "aneurysmal bone cyst" was used and described by Jaffe and Lichtenstein in 1942 [9]. In 1958, the first clinical case of an aneurysmal bone cyst of the jaw was described by Bernier and Bhaskar [3]. Most ABCs of the jaws seem to occur in adolescents, predominantly those younger than 20 years and appear to have a slight predilection for females [10]. The lesion is more commonly seen in the mandible than maxilla (3:1), and, in particular, the ramus and posterior body account for 51.7% of the lesions. Only 11 cases have been reported so far in peer-reviewed English literature affecting the condyle in children less than 12 years of age.

In our report, the mean age of presentation was 9.67 years with two males and one female patient. To the best of the authors' knowledge, this is the first report of a case series of aneurysmal bone cyst arising in the temporomandibular area affecting exclusively the paediatric population. Table 1 presents the gathered data from the published case reports in paediatric patients and compares it with our cases.

S. No.	Authors	Age	Sex	Side	Size of the lesion	Symptoms	Treatment	Reconstruction	Recurrence	Follow Up (Months)
1.	Motamedi., *et al*. [13]	11	F	Rt	Not Stated	Pain Pre-auricular Swelling	Curettage	None	None	12
2.	Gadre and Zubairy., *et al*. [18]	12	F	Lt.	1.5X 2 cm	Pain, Pre-auricular swelling, Joint pain, Limited mouth opening	Resection	Costochondral Graft	None	60
3.	Pelo., *et al*. [19]	10	M	Rt	Not Stated	Pre-auricular Swelling	Resection	None	None	36
4.	Fernandez., *et al*. [20]	10	F	Lt.	Not Stated	Pre-auricular Swelling	Embolization	None	Yes	Not Stated
	Our Case Series									
5.1		11	F	Rt.	3.3. X 3.2	Pre-auricular swelling	Resection	Costochondral Graft	None	21
5.2		8	F	Lt.	4.2 X 3.6	Pre-auricular swelling	Resection	Costochondral Graft	None	14
5.3		10	M	Lt.	4.5 X 3.7	Pre-auricular swelling, Joint Pain	Resection	Costochondral Graft	None	12

Table 1: *Previously reported paediatric cases of patients with condylar aneurysmal bone cyst.*

The aetiopathogenesis of the ABC is controversial and has been a subject of ongoing research ever since the pathology was first described [11]. It remains unclear whether the lesion is primary or secondary to a pre-existing osseous lesion. Some authors have described it as a congenital lesion; while others have claimed that its origin lies in trauma. 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. Other authors argue that it emanates from degeneration of a pre-existing lesion such as central giant cell granuloma, fibrous dysplasia, haemangioma, eosinophilic granuloma, ossifying fibroma, or chondroblastoma, among others [2].

The clinical signs and symptoms of the condylar ABC are diverse and non-specific [12-15]. It usually manifests as an inflammation of the soft tissues due to expansion of the cortical bone, and later as the development of facial asymmetry and malocclusion. In the most aggressive cases, considerable bone destruction takes place, even invading the soft tissues. Other associated symptoms may include pain, paraesthesia, mobility, migration, or resorption of the involved teeth, depending on the extent of the lesion. As with clinical features, the radiological manifestations of these lesions are also variable. The typical radiographic picture is that of a unilocular radiolucent lesion or multilocular expanse, with cortical bone destruction and an internal trabeculated pattern. The multiloculated pattern endows the radiograph with a characteristic "honeycomb," "soap bubble," or "moth-eaten" appearance. CT is the imaging 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 imaging (MRI) helps better to visualize soft tissue affectation and the presence of liquid content. The very non-specific clinical symptoms, as well as the radiological appearance, makes the initial diagnosis of aneurysmal bone cysts difficult. Other lesions with a similar presentation such as ameloblastomas, ossifying fibroma, giant-cell granulomas or keratocyst have to be taken into account in the differential diagnosis for aneurysmal bone cysts and should be excluded with histopathological evaluation.

Treatment of ABC depends on the size and localisation of the cyst, the clinical presentation and the age of the patient. According to Zadik., *et al*. [16], condylar ABCs show a recurrence rate of 60% after curettage and 20% after resection. A simple classification of aneurysmal bone cysts in three stages according to radiological and clinical aspects was introduced by Capanna., *et al*. in 1985 [17]:

1. The inactive stage presents with complete periosteal and sclerotic borders
2. The active stage shows incomplete periosteal borders with defined margins
3. The third, aggressive, stage is described as a uniform osteolysis with diffuse boarders of the lesion.

Active and aggressive cysts tend to recur, whereas the inactive cysts do not show any proliferation. Therefore, Inactive cysts may be treated by curettage, whereas active and aggressive lesions with painful symptoms, evidence of progression and osteolysis should be resected. All cases (3 out of 3; 100%) in our series presented in the active stage, necessitating radical management. In our cases, resection was followed by immediate reconstruction with a costo-chondral graft, so chosen because of its autogenous origin and potential for growth, a prerequisite in paediatric cases. Also, with total joint prosthesis being contraindicated in the developing mandible and the results from distraction osteogenesis and other reconstruction being encouraging yet largely unrefined, a better alternative has yet to be identified.

All recurrences in the literature have been reported within 12 months of the surgery [18-20]. Thus, a close post-operative follow-up of at least 12 months is warranted. All our cases revealed no evidence of recurrence of the disease or graft overgrowth at any stage of follow-up, with achievement of good symmetry with normal mandibular function.

The types of surgical treatments for Condylar ABCs vary from curettage to resection without reconstruction (low-condylectomy), resection (total condylectomy) with TMJ reconstruction (autogenous/alloplastic). Although previous studies have reviewed various modes of treatments and have supported the efficacy of each of them, none address the indications of such treatments in an algorithmic fashion. In this study, we reviewed a series of cases which were surgically treated and proposed a treatment algorithm (Figure 6) based on initial presentation, age, location of the mass, morphology of the mass, and the degree of dentofacial deformity created.

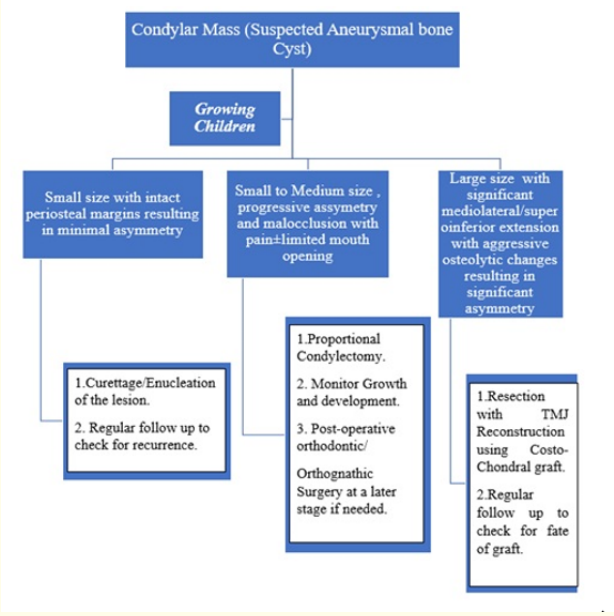

Figure 6

Conclusion

ABC of the jaws is an enigmatic pseudocyst due to its variable presentations. The diagnosis should be based on a combination of clinical, radiographical and histopathological features. The anatomical location and proximity of the condylar ABC to the temporomandibular joint could have long-term functional implications. Therefore, proper anatomical relationships should be accurately re-established.

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Conflict of Interest

The authors declare that they have no conflict of interest.

Ethical Approval

All procedures performed in study involving human participants were in accorda1

ce with the ethical standards of the institutional research committee and with the 1964 Helsinki declaration.

Informed Consent

Informed consent was obtained from patient included in this study.

Bibliography

1. Schajowicz F, Schajowicz F. Histological Classification of Bone Tumours. In: Histological Typing of Bone Tumours. Springer Berlin Heidelberg; 1993:3-6.
2. Perrotti V, Rubini C, Fioroni M, Piattelli A. Solid aneurysmal bone cyst of the mandible. Int J Pediatr Otorhinolaryngol. 2004;68(10):1339-1344.
3. Bernier JL, Bhaskar SN. Aneurysmal bone cysts of the mandible. Oral Surgery, Oral Med Oral Pathol. 1958;11(9):1018-1028.
4. Motamedi MHK, Khodayari A. Aneurysmal bone cyst mimicking a malignancy. J Oral Maxillofac Surg. 1993;51(6):691-695.
5. Trent C, Byl FM. Aneurysmal bone cyst of the mandible. Ann Otol Rhinol Laryngol. 1993;102(12):917-924.
6. Hardee PS, Whear NM, Morgan PR. Aneurysmal bone cyst of the maxilla--an association with tooth resorption. J Craniomaxillofac Surg. 20(6):266-269.
7. Kalantar Motamedi MH, Yazdi E. Aneurysmal bone cyst of the jaws: Analysis of 11 cases. J Oral Maxillofac Surg. 1994;52(5):471-475.
8. Van Arsdale WW. Ossifying Hæmatoma. Ann Surg. 1893;18(1):8-17.
9. Jaffe H. Solitary unicameral bone cyst: with emphasis on the roentgen picture, the pathologic appearance and the pathogenesis. Arch Surg. 1942;44(6):1004-1025.

10. Jones RS, Dillon J. Nonodontogenic Cysts of the Jaws and Treatment in the Pediatric Population. Oral Maxillofac Surg Clin North Am. 2016;28(1):31-44.

11. Cottalorda J, Bourelle S. Modern concepts of primary aneurysmal bone cyst. Arch Orthop Trauma Surg. 2007;127(2):105-114.

12. Rapidis AD, Vallianatou D, Apostolidis C, Lagogiannis G. Large lytic lesion of the ascending ramus, the condyle, and the infratemporal region. J Oral Maxillofac Surg. 2004;62(8):996-1001.

13. Motamedi MHK. Destructive aneurysmal bone cyst of the mandibular condyle: Report of a case and review of the literature. J Oral Maxillofac Surg. 2002;60(11):1357-1361.

14. Ma J, Calleja L-A, Luis J, et al. Aneurysmal bone cyst of the mandible: Case presentation and review of the literature. Med Oral Patol Oral Cir Bucal. 2007;12:E401-E403.

15. Newman L. Aneurysmal bone cyst-A lesion in the mandibular ramus. Br J Oral Maxillofac Surg. 1987;25(1):74-78.

16. Zadik Y, Aktaş A, Drucker S, Nitzan DW. Aneurysmal bone cyst of mandibular condyle: A case report and review of the literature. J Cranio-Maxillofacial Surg. 2012;40(8):e243-e248.

17. Capanna R, Bettelli G, Biagini R, Ruggieri P, Bertoni F, Campanacci M. Aneurysmal cysts of long bones. Ital J Orthop Traumatol. 1985;11(4):409-417.

18. Gadre KS, Zubairy RA. Aneurysmal bone cyst of the mandibular condyle: report of a case. J Oral Maxillofac Surg. 2000;58(4):439-443.

19. Pelo S, Gasparini G, Boniello R, Moro A, Amoroso PF. Aneurysmal bone cyst located in the mandibular condyle. Head Face Med. 2009;5(1):8.

20. Marín Fernández AB, García Medina B, Martínez Plaza A, Aguilar-Salvatierra A, Gómez-Moreno G. Aneurysmal bone cyst of the mandible affecting the articular condyle: a case report. Clin Case Reports. 2016;4(12):1175-1180.

Volume 5 Issue 5 May 2022