

*Case report***Rare mandibular condylar pathology and new surgical approach: intraoral approach for resection and reconstruction of aneurysmal bone cyst, a case report****Mojtaba Azadbakht¹, Amir Yari^{1,*}, Paniz Fasih², Mohammad Nomani¹ and Amirali Asadi¹**

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Abstract: The case of a 27-year-old male with a destructive aneurysmal bone cyst of the right mandibular condyle, coronoid process and ascending ramus is presented. Surgery revealed an eggshell-thin, partly perforated cortices. Intraoral approach was used for a complete resection of mandibular condyle, coronoid process and affected ascending mandibular ramus. Curettage of the adjacent soft tissues performed; no extra oral incision was used. In this case following resection, the defect was primarily reconstructed with prefabricated reconstruction plate with condylar head to maintain functional mandibular ramus height, avoid malocclusion and asymmetry. Post-operative examination and radiographic images revealed good symmetry and perfect occlusion as well as almost normal mandibular function. Intraoral approach can be used to access to the mandibular condyle region and ascending ramus with no risk to facial nerve and skin scar so cosmetically more favorable.

Keywords: aneurysmal bone cyst; mandibular condyle; intraoral approach

1. Introduction

In 1950, Jaffe and Lichtenstein first described clinical, radiographic, and pathologic characteristics of aneurysmal bone cyst (ABC) independently [1,2]. ABC is a benign pseudo cystic osseous lesion and act as a osteolytic and expansile lesion characterized by a fibrous connective tissue

stroma, osteoclast-like multinucleated giant cells and blood-filled spaces separated by fibrous septa without any endothelial lining [3–5].

ABC commonly occurs in the long bones (50%) and vertebral column (20%) although any bone may be affected [6,7]. Development of the lesion in the craniofacial region is rare, and the mandible is more often affected than the maxilla. In the jaws, two-thirds are located in mandible and one-third in the maxilla (reported cases in the mandible contain: body of the mandible 40%, the ramus 30% and the angle 19%) [6,8,9]. Mandibular condyle involvement is extremely rare [10]. It usually occurs in patients younger than 20 years of age, and shows no gender predilection. Aneurysmal bone cysts constitute about 1.5% of all nonodontogenic cyst-like lesions in the jaws [11].

Different surgical approaches to the mandibular condyle and ramus have been described. A survey found that the preauricular and submandibular approaches are preferred respectively for reconstruction of TMJ region [12]. The aim of this manuscript is to describe a rare pathologic lesion in mandibular condyle and intraoral approach for resection and reconstruction of mandibular condyle and ascending ramus affected by ABC.

The ABC is a benign solitary osseous lesion. The etiology of the ABC remains elusive. There are various theories regarding the pathogenesis of the ABC like trauma, subperiosteal intraosseous hematoma, and reactive vascular malformation [13]. In recent years, chromosomal alterations of segments 17p and 16q have been described suggesting a neoplastic origin of the lesion. The incidence of the solid type of ABC has been reported as 5% to 7.5% [14]. The lesion usually occurs in young patients below the age of 20 years. No gender predilection is seen, and the most common site in the maxillofacial skeleton is the mandible (predominantly in the posterior region), followed by the maxilla. The mandibular condyle is rarely affected [6,9,11,14]. Histopathologically, it consists of two main variants: The vascular variant includes about 95% of total cases of ABC while the solid type is rare and consists of only 5–8% of reported cases [9]. ABC can be associated with other pathologic lesion which giant cell granuloma is the most common. Other reported associated lesions consist of brown tumor, chondroblastomas, osteoblastoma, fibrous dysplasia, hemangioma and ossifying fibroma [7].

2. Case report

An otherwise healthy 27-year-old male referred to Shariati Hospital, with a 1-year history of pain in the right preauricular region and swelling over the same region within past 6-months.

Clinical examination revealed a smooth and diffuse swelling approximately 4 cm × 5 cm in size in right preauricular region (Figure 1). More examinations also revealed presence of moderate tenderness, minimal decrease in maximum mouth opening and mandibular deviation while mouth opening. Submandibular and cervical lymph nodes were not palpable. No sensory impairment, facial nerve and skin were completely intact. Blood tests were unremarkable.

On radiographic examination, there was a large multinucleated radiolucent lesion with scalloped borders in the area of right mandibular condyle in panoramic view (Figure 2). The initial differential diagnoses proposed were Ameloblastoma and Keratocystic odontogenic tumor (KOT). Further imaging with computed tomography (CT) and magnetic resonance imaging (MRI) revealed a 4.8 cm × 3.7 cm sized expanded, cystic and fluid filled lesion with septa of the right mandibular condyle suggesting an ABC (Figure 3,4). Coronal CT scans revealed an intrabony lesion with near symmetrical expansion of the medial and lateral cortices (Figure 5).



Figure 1. Swelling and asymmetry on right side of the face.



Figure 2. Large multinucleated radiolucent lesion with scalloped borders in the area of right mandibular condyle.

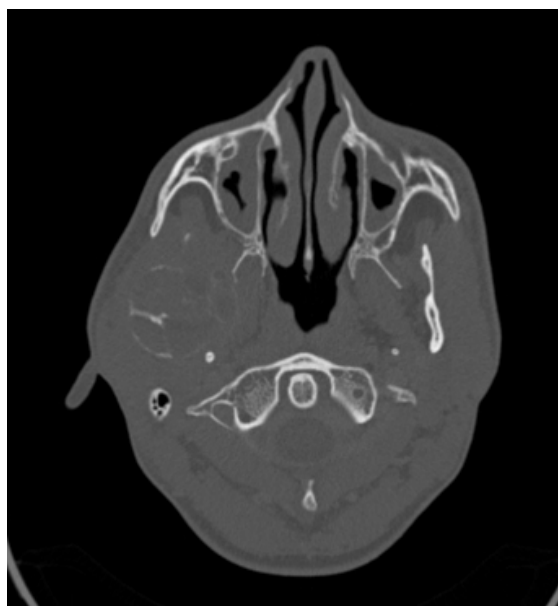


Figure 3. Cystic lesion with septa of the right mandibular condyle.

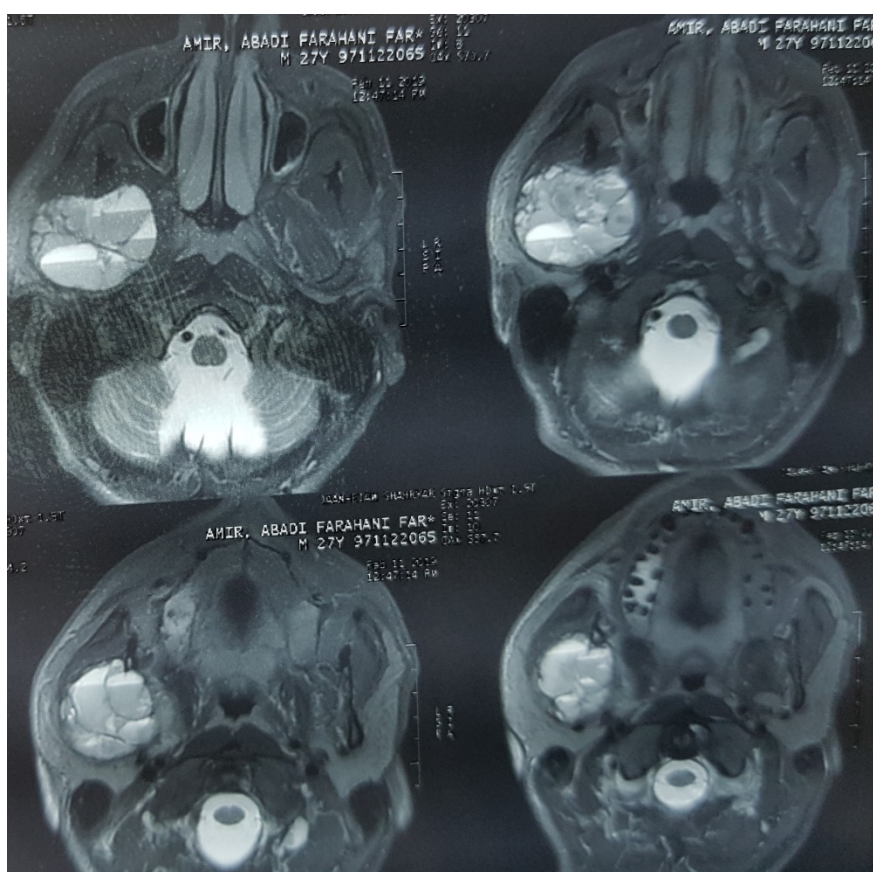


Figure 4. Cystic and fluid filled lesion with septa of the right mandibular condyle.



Figure 5. Intrabony lesion with near symmetrical expansion of the medial and lateral cortices.

Aspiration was performed using a large-bore needle, and blood was readily aspirated. Fine needle aspiration and core needle biopsy were performed before planning surgery and histopathologic examination showed a benign tumor containing fibrous tissue and giant cells.

Under general anesthesia via nasotracheal intubation and after routine patient preparation, Intraoral approach was used for a complete resection of mandibular condyle, coronoid process and affected ascending mandibular ramus (Figure 6). The articular disc was not invaded so was left in place. The lateral cortex of mandibular ramus was egg-shell thin and perforated partly. Brown and relatively vascular granular tissues were curetted out of the bone (Figure 7). The medial cortex was also expanded and perforated. Bleeding was mild during the procedure. Following surgery, the patient developed slight paresthesia of the mandibular nerve, which resolved several weeks later. The defect was primarily reconstructed with prefabricated reconstruction plate with condylar head to maintain functional mandibular ramus height, avoid malocclusion and asymmetry. A complete stereolithographic model of the patient's mandible was made. A 3D model of the mandible was constructed using the mirror image technique for pre-operative bending and shaping of the titanium plate before operation (Figure 8). The patient's recovery was uneventful. Active mouth opening for patient started immediately after removal of intermaxillary fixation after 3 weeks. Histopathological examination of resected lesion revealed a connective tissue stroma containing cellular fibrous tissue with cystic spaces and osteoclast-like multinucleated giant cells with hemorrhage in lumina of cysts and the final pathology report confirmed the diagnosis of an ABC (Figure 9).

Post-operative examination and radiographic images revealed ideal symmetry and satisfactory occlusion as well as almost normal mandibular function except from slight deviation to the right upon opening after 4 weeks (Figure 10). There was no evidence of any recurrence after 1 year of follow-up.

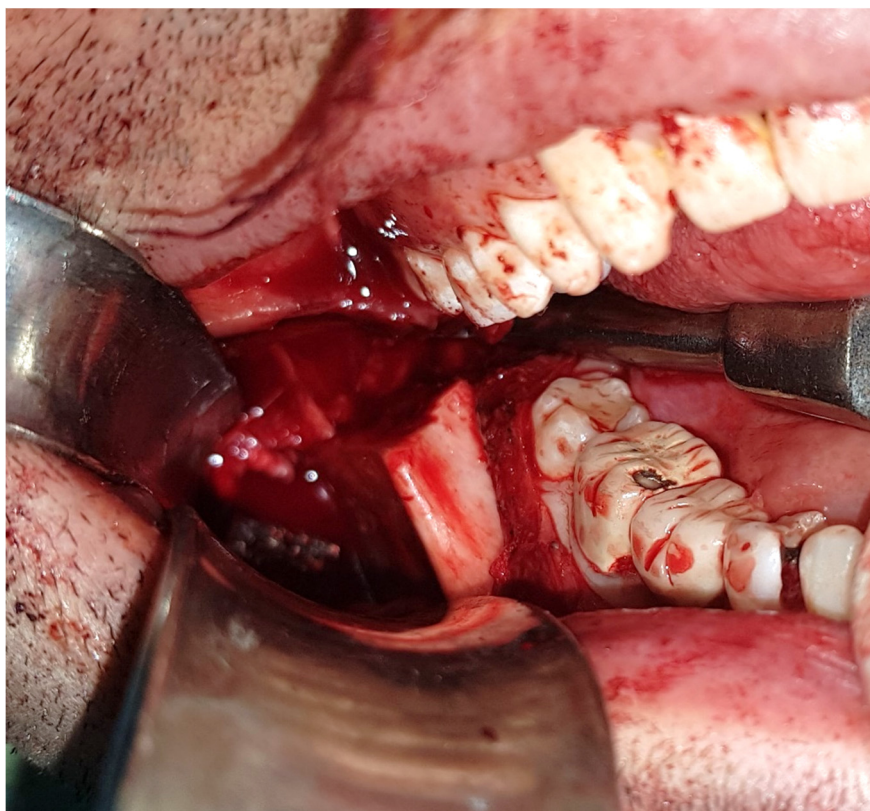


Figure 6. Intraoral approach was used for access to the lesion.

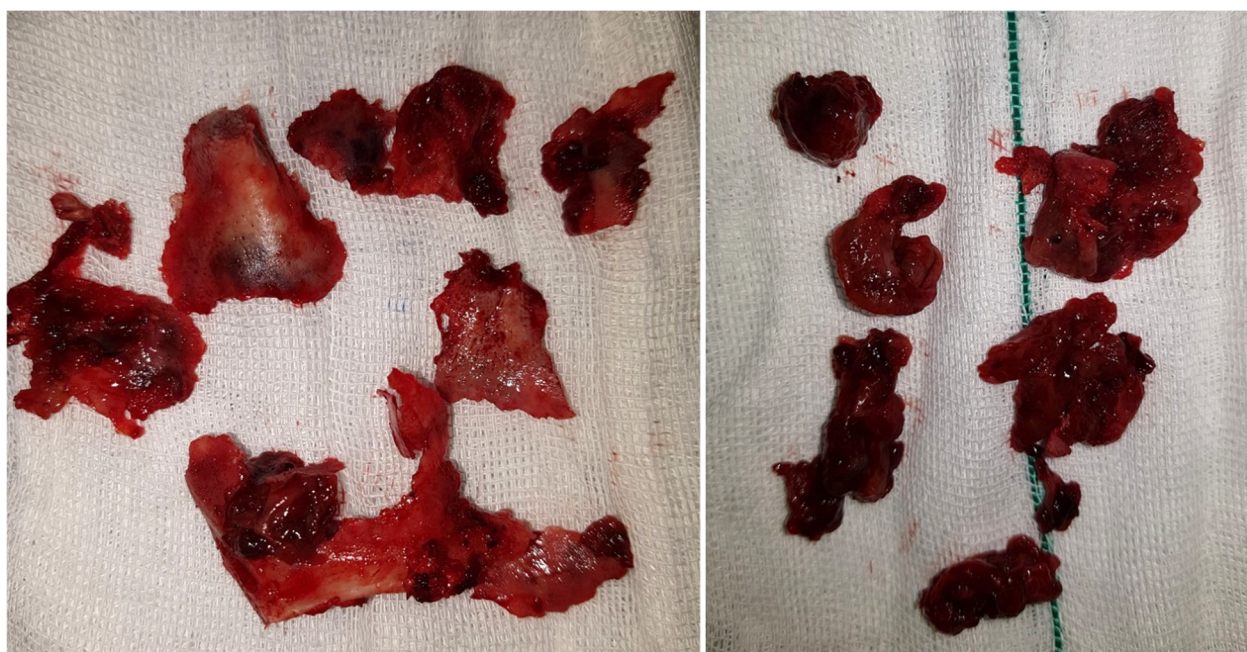


Figure 7. Brown and relatively vascular granular tissues were curetted out of the bone.



Figure 8. A 3D model was constructed using the mirror image technique for pre-operative bending of the titanium plate.

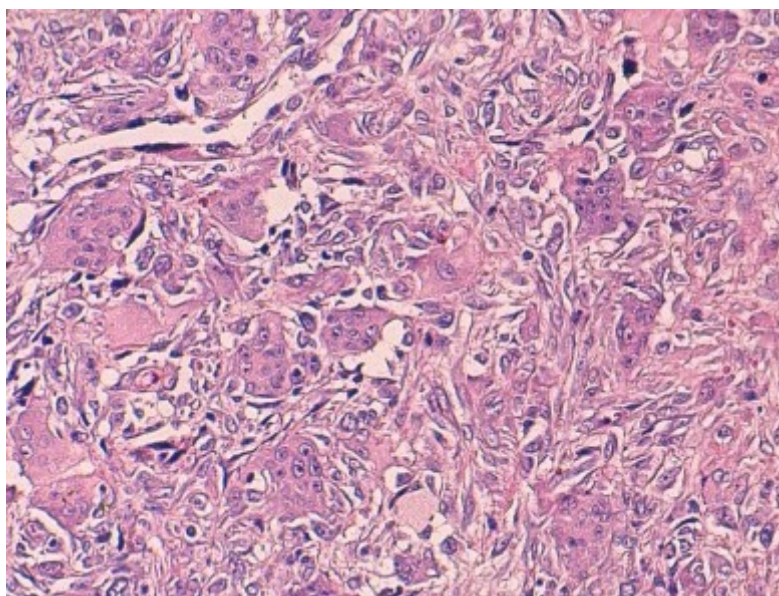


Figure 9. Histopathological specimen: connective tissue stroma containing cellular fibrous tissue with cystic spaces and osteoclast-like multinucleated giant cells.



Figure 10. Post-operative radiographic images revealed ideal symmetry and satisfactory occlusion.

3. Discussion

The main clinical manifestation is swelling and generally signs and symptoms are nonspecific and do not contribute to the clinical diagnosis [15]. The associated pain of condylar ABC might be due to secondary temporomandibular dysfunction [16].

In the present case also, there was presence of swelling, asymmetry and associated pain secondary to temporomandibular joint dysfunction. CT scan showing different fluid levels and histologically, blood-filled spaces within connective tissue stroma, and osteoclastic multinucleated giant cells which is pathognomonic of ABC together has led to the definitive diagnosis of ABC.

Table 1 is a comparison of the features of condylar ABC and noncondylar ABC of the mandible [8].

Table 1. Comparison of clinical features of condylar and noncondylar-ABC.

Features	Condylar ABC	Noncondylar ABC
Mean age	17	20
Swelling and asymmetry	100%	74%
Pain and tenderness	72%	43%
Recurrence after curettage	60%	15%
Recurrence after resection	20%	12%

A variety of treatments have been recommended for ABC, ranging from long-term observation which may lead to spontaneous regression to simple curettage, cryotherapy, excision, block resection and bone grafting, therapeutic embolization, open packing, intralesional calcitonin and methylprednisolone injection. Simple curettage is associated with high recurrence rates varying from 21 to 50% [17–19]. The present case was treated by resection and reconstruction since the lesion size was large and the pattern was multilocular with bony septa. There was no evidence of any recurrence after 1 year of follow-up. In this case intraoral approach was used without any extraoral incision. Intraoral approach can be used to access to the mandibular condyle region and ascending ramus with no risk to facial nerve and skin scar so cosmetically more favorable. The most commonly used

approach to the temporomandibular joint region is preauricular incision; disadvantages include scar formation, loss of sensation, facial nerve damage and Frey's syndrome [20].

Reconstruction of the mandible remains a challenge in surgical therapy. In the present case data from CT scan images were used to create a stereolithographic model of the mandible using a rapid 3D printer. A mandibular reconstruction plate was then accurately and precisely bent to fit the contours of the defect preoperatively. Precontouring significantly shortened the operation time. Precise adaptation of the plate and excellent symmetry was achieved in a considerably short operation time. Other benefits which should be mentioned include better understanding of bone morphology; which provides for more accurate and easier planning of preoperative plate bending, decreased exposure time to general anesthesia, decreased blood loss and shorter wound exposure time [21].

By analyzing the results of 7 cases of reconstruction of condylar ABC since 2000 (Table 2) [11,15,22–24] we found that, 61% of cases were reconstructed by costochondral graft, while 25% cases has gone for condylar prosthesis as an reconstructive option. As we have mentioned before, prefabricated reconstruction plate with condylar head was used as reconstructive option to maintain functional mandibular ramus height, avoid malocclusion and asymmetry (A 3D model of the mandible was constructed using the mirror image technique for pre-operative bending and shaping of the titanium plate). Post-operative examination and radiographic images revealed good symmetry and normal occlusion as well as almost normal mandibular function.

Table 2. Reconstruction review of ABC condyle resected cases.

Author's	Reconstruction option
Gadre et al. (2000)	Costochondral graft
Rapidis et al. (2004)	Costochondral graft
Pelo et al. (2009)	Bionator functional therapy for growth stimulation
Ettl et al. (2009)	Condylar head add on system
Zadik et al. (2012)	Costochondral graft
Ziang et al. (2013)	Costochondral graft
Indu Bhusan Kar et al. (2015)	Condylar head add on system

4. Conclusion

We present the clinical manifestation and management of an aneurysmal bone cyst lesion in the mandibular condyle of a 27-year-old man. The ABC lesion in the mandibular condyle is extremely rare and there is a paucity of condylar-ABC reports in the literature. Surgical resection and immediate reconstruction should be considered as treatment of choice for condylar-ABC. Prefabricated reconstruction plate with condylar head has shown satisfactory results and should be considered as a preferred primary reconstructive option, reserving the autograft for recurrence cases. Intraoral approach can be used for resection and reconstruction of mandibular ramus and condyle with avoiding obvious disadvantages of extraoral approaches like skin scars and risk of facial nerve damage. Close post-operative follow up, for at least 1 year is recommended for mandibular ABC especially when the lesion is located in the condyle.

Conflict of interest

The authors declare no conflict of interest.

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