



Multilocular radiolucency of the mandibular condyle in a 19-year-old woman

I-Yueh Huang, DDS, MS,^{a,b} Yi-Lun Shih, DDS, MS,^a Ching-Wei Shu, DDS, MS,^a and Yuk-Kwan Chen, DDS, MS,^{b,c} Kaohsiung, Taiwan
 KAOHSIUNG MEDICAL UNIVERSITY
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A 19-year-old woman presented to the orthodontic department of our institution for evaluation of maxillary excess and mandibular deficiency. On routine cephalometric and panoramic images, an asymptomatic multilocular radiolucent lesion was incidentally noted in the left mandibular condylar area (Fig. 1). The patient was subsequently referred to oral and maxillofacial surgery for a consultation regarding both orthognathic surgery and treatment of the condylar lesion. After the consultation, the patient elected to undergo presurgical orthodontic treatment.

Follow-up imaging of the condylar lesion (Fig. 2) was performed 7 months after the first images were obtained. Although the lesion was found to be approximately the same size, the orthognathic surgeon referred the patient to an oral and maxillofacial surgeon who specializes in diseases of the temporomandibular joint (TMJ) for further examination.

On examination by the second surgeon, the patient denied any history of trauma. The patient described the left TMJ as "tight," and mild crepitus was noted. These symptoms were noteworthy because they had not been reported previously. The patient denied any pulsation, paresthesia, or pain. Joint sounds from the bilateral

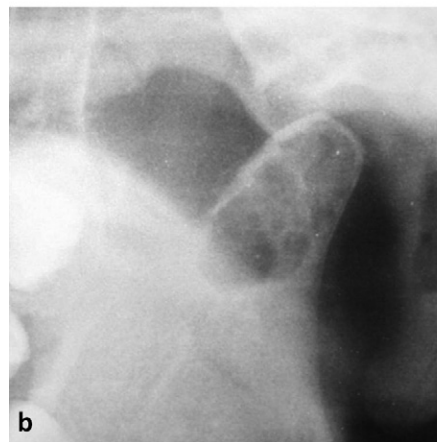
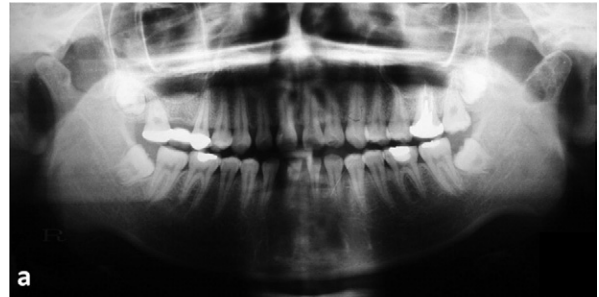


Fig. 1. a, Panoramic image taken for orthodontic evaluation in February 2009 showing a multilocular radiolucent lesion involving the left condylar head and neck. b, Cropped close-up view of the left condylar lesion.

^aDivision of Oral and Maxillofacial Surgery, Department of Dentistry, Kaohsiung Medical University, Chung-Ho Memorial Hospital.

^bSchool of Dentistry, College of Dental Medicine.

^cDivision of Oral Pathology and Diagnosis, Department of Dentistry, Kaohsiung Medical University, Chung-Ho Memorial Hospital.

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TMJs could not be detected by stethoscope. A normal appearance of the skin and oral mucosa was noted. Interincisal opening was measured to be ~54 mm. A

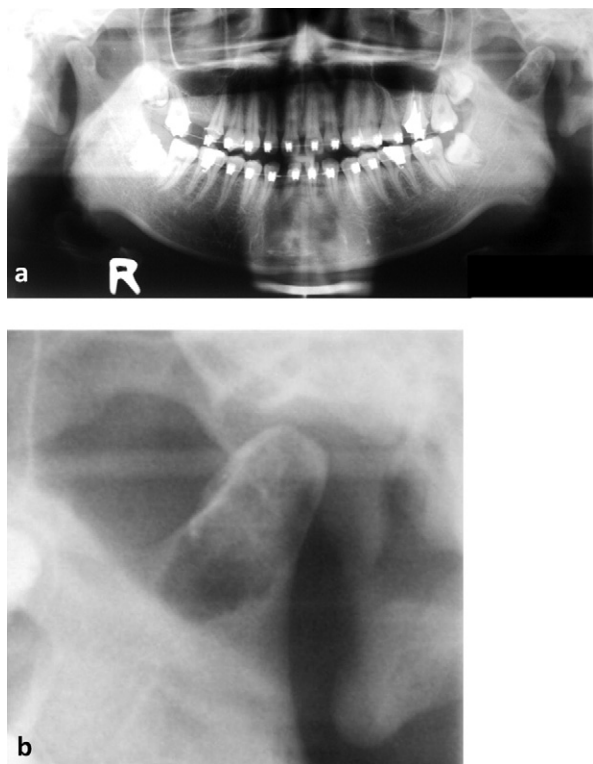


Fig. 2. a, Panoramic image taken 7 months after the image in Fig. 1, revealing a lesion of similar size with slight expansion. b, Cropped close-up view of the left condylar lesion.

3-dimensional computerized tomographic (CT) scan revealed an osteolytic lesion without bony expansion involving the outer aspect of the left condylar neck (Fig. 3, a) and a deformity with an osteolytic lesion at the inner aspect of the condylar head (Fig. 3, b).

DIFFERENTIAL DIAGNOSES

The mandibular condyle is a region in which tumor/tumor-like lesions rarely form; osteoma, osteochondroma, chondroma, and synovial chondromatosis being the most common pathologic entities.¹ However, these lesions are predominantly radiopaque rather than radiolucent lesions. Therefore, our differential diagnoses of this well defined multilocular radiolucent lesion of the left mandibular condyle included benign nonodontogenic lesions, such as aneurysmal bone cyst (ABC), hemangioma, central giant cell granuloma (CGCG), desmoplastic fibroma, benign fibrous histiocytoma, and chondromyxoid fibroma. Although odontogenic cysts and benign odontogenic neoplasms, such as keratocystic odontogenic tumor, glandular odontogenic cyst, and ameloblastoma, are relatively common causes of multilocular radiolucent lesions of the mandible, these lesions were considered to be less likely in the present

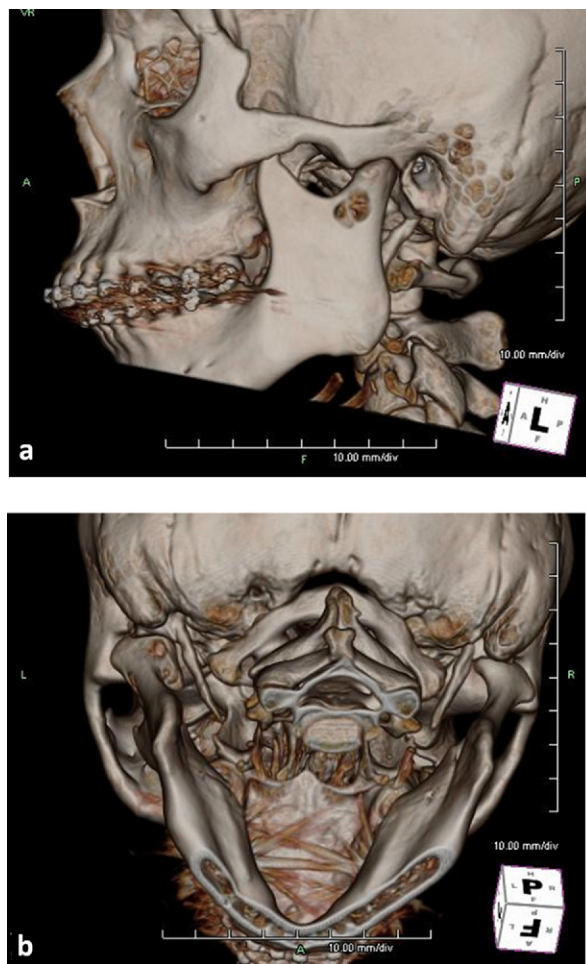


Fig. 3. Three-dimensional computerized tomographic scan demonstrates an osteolytic lesion at the outer side of the left condylar neck (a) and deformity of the inner side of the left condylar head (b).

case because it was restricted to the mandibular condyle, with no involvement of the tooth-bearing areas of the mandible. Odontogenic cysts and tumors, such as keratocystic odontogenic tumor or ameloblastoma may involve the TMJ, but these lesions are frequently associated with an impacted tooth and typically originate from the mandibular body or angle, whence they may extend to the mandibular condyle.² However, the odontogenic myxoma has been reported in the nontooth-bearing area of the mandible.³

ABC of the jaw is a pseudocystic lesion that may be of primary origin or a secondary phenomenon in a preexisting bone lesion (e.g., CGCG).⁴ A hemodynamic disorder and arteriovenous malformations are hypothesized to enhance the intraosseous venous pressure, leading to enlargement of the vascular bed and resulting in bone resorption and substitution by connec-

tive tissue and reactive osteoid.⁴ The clinical presentation of an ABC in the jaws can include a firm swelling with or without tenderness, progressive enlargement, and occasionally perforation of the bony cortex.⁴ Unilocular or multilocular radiolucent lesions with thinning of bony cortices and with well defined or diffuse margins have been reported.⁴ The average age of occurrence is 13 years, and 80% of patients are <20 years old, with no gender predilection.⁴ Although the exact relationship between trauma and ABC has yet to be confirmed, 50%-70% of patients with an ABC may have a history of trauma.⁵ The present case was largely compatible with this clinical information, although no history of trauma was noted. To our knowledge, 6 cases of ABC originating in the mandibular condyle have been reported in the English-language literature,⁶⁻¹⁰ 3 of which presented as multilocular lesions.^{6,7,9} Therefore, we considered an ABC to be the most likely possibility in our differential diagnosis.

Because imaging of central hemangioma usually presents as a multilocular radiolucent shadow with a honeycomb or soap bubble appearance,¹¹ this vascular neoplasm was included in our differential diagnosis. Patients with central hemangioma of the jaw bone are usually asymptomatic. However, symptoms, such as pain, swelling, discomfort, pulsation, tooth mobility and slow oozing from the gingiva, especially in high-flow vascular lesions, may be present.¹²

Another radiolucent lesion found typically in children and young adults that was considered is CGCG. Most cases of CGCG occur before the patient is 30 years of age, and it is found slightly more often in female patients. Radiologically, this lesion may appear as a well defined unilocular or multilocular radiolucency with undulating borders.¹³ At least 4 cases of CGCG originating in the mandibular condyle¹³ have been documented in the English-language literature, 1 of which presented as a multilocular radiolucency on panoramic imaging.¹⁴

Though uncommon, desmoplastic fibroma, benign fibrous histiocytoma, and chondromyxoid fibroma were also considered as possibilities.

Desmoplastic fibroma is a myofibroblastic neoplasm, with about 84% of previously reported jaw lesions affecting the mandible, particularly the posterior region. Radiologically, this lesion may manifest as an ill- or well-defined radiolucency, usually with a multilocular pattern. Desmoplastic fibroma that arises intraosseously can perforate bone (as noted in the current lesion). On the other hand, desmoplastic fibroma that occurs in soft tissue can also infiltrate underlying bone. Moreover, most cases of desmoplastic fibroma have occurred in patients under 30 years of age with no significant sex predilection. The majority cases have presented as

asymptomatic swellings; however, trismus and tooth mobility may infrequently be noted.¹⁵

At least 1 case of a benign fibrous histiocytoma primarily affecting the mandibular condyle has been reported.¹⁶

Chondromyxoid fibroma is a slow-growing benign bone neoplasm that is characterized by chondroid and myxoid differentiation.¹⁷ According to a review of the English-language literature, 22 cases with an equal gender distribution have been reported to affect the jaws with mandibular predominance.¹⁷ The most common complaint is pain or swelling in the involved area,¹⁷ a feature not noted in the present case. The radiologic features are those of a well defined radiolucency with sclerotic or scalloped margins. The outline may appear irregular, and the cortex of involved bone may be expanded or thinned. Radiologic evidence of calcification is occasionally apparent.¹⁷ This lesion is most common among patients in the second or third decades of life.¹⁷

Finally, owing to the radiographic presentation as a multilocular radiolucency, odontogenic myxoma was also considered. Radiologically, a multilocular presentation is more commonly noted, with cystic-like areas of variable radiolucency separated by many bony trabeculae, usually of a well defined nature.¹⁸ To date, 1 well confirmed myxoma of the mandibular condyle has been reported.³ Clinically, the mean age of presentation of myxoma is between the second and fourth decades with a higher female prevalence.¹⁸

DIAGNOSIS AND MANAGEMENT

Owing to the fact that the expansile osteolytic lesion involved most of the condyle and to concern about the possibility of recurrence if conservative curettage were performed, complete surgical excision was regarded to be the treatment of choice in the present case. Therefore, the patient was advised to undergo condylectomy and rib costochondral grafting. Under general anesthesia, a retromandibular incision was made to reflect the masseter muscle and to expose the right mandibular angle, ramus, sigmoid notch, and condylar head. Aspiration was performed at the outer surface of the condylar neck but no blood was noted. Subcondylectomy was performed by reciprocal saw, and the capsule was stripped off. When the condylar head containing the lesion was removed, no excessive bleeding was noted. Furthermore, the lesion was not found to have invaded the adjacent soft tissue, and the disk was preserved. For reconstruction of the defect, a costochondral graft was harvested from the right seventh rib by a cardiothoracic surgeon, and ~5 cm of bone with 5 mm of cartilage was fixed to the left ramus via 3 transosseous wires. A drainage tube was subsequently inserted. Intermaxillary

Fig. 4. Decalcified section of the surgical specimen revealing bony trabeculae separated by multiple thin-walled, dilated, blood-filled vascular spaces lined by flattened endothelial cells (hematoxylin and eosin stain, $\times 200$).

fixation was placed for 6 weeks, and mouth-opening exercises (MOE) were prescribed. After 3 weeks of MOE, the interincisal distance was measured to be 43.5 mm. Very mild left facial nerve weakness was noted after surgery, which had completely recovered 3 months after surgery.

The bony surgical specimen was sent for histopathologic examination. Routine hematoxylin and eosin staining of the decalcified specimen demonstrated mature bony trabeculae separated by multiple thin-walled, dilated, blood-filled vascular spaces lined by flattened endothelial cells (Fig. 4). No abnormal mitotic activity was noted. The final microscopic diagnosis was vascular malformation of the left mandibular condyle.

DISCUSSION

Due to the rarity of vascular lesions involving the mandibular condyle, most of the available literature consists of case reports. We were able to identify only 7 cases of intraosseous vascular malformation/hemangioma involving the mandibular condyle.¹⁹⁻²⁵ The characteristics and management of vascular condylar lesions remain uncertain. In those cases in which the age of the patient was supplied,^{19,23-25} including the present case, a range of 19 to 54 years was noted, our patient being the youngest. Pain was the most commonly reported symptom (6 cases),²⁰⁻²⁵ and swelling was noted in 2 cases.^{24,25} Including the present case, 2 cases¹⁹ were asymptomatic. Two cases also involved the ramus,^{23,24} whereas 3 cases, including our case, were limited to the condylar head and condylar neck.^{19,25} Histopathologically, 5 cases were diagnosed

as hemangioma²⁰⁻²⁴; the other 3 cases, including ours, were diagnosed as vascular malformation.^{19,25}

The treatment for mandibular condylar vascular malformation/hemangioma varies depending on the size, location, and vascular flow rate of the lesion. In most reports, excessive bleeding was not noted during surgery. Condylectomy has been reported using different reconstruction methods, including no grafting (3 cases),²⁰⁻²² sliding vertical ramal osteotomy (1 case),¹⁹ and reconstruction with a TMJ prosthesis (3 cases).²³⁻²⁵ The present report is, to our knowledge, the first case of mandibular condylar vascular malformation treated by surgical excision and reconstruction by rib costochondral grafting.

According to Cheng et al.,¹² magnetic resonance imaging should be performed first to define the extent of the lesion as well as its relationship with surrounding structures. Moreover, when a vascular lesion is confirmed, angiography should be performed before surgical intervention to evaluate whether the flow of the lesion is high or low, especially when presented with a soap bubble or honeycomb osteolytic lesion with poorly defined and demarcated borders. If such an examination is unavailable, the patient should be followed closely, with periodic examination by conventional radiography. This is because early recognition of the lesion is important to prevent uncontrolled bleeding as well as to avoid permitting a lesion to grow to the extent that it becomes inoperable.¹²

In conclusion, primary intraosseous vascular lesions affecting primarily the mandibular condylar region are extremely rare. In the present case, the patient was asymptomatic, and a CT scan did not suggest a lesion of possible vascular origin.

REFERENCES

1. Warner BF, Luna MA, Robert-Newland T. Temporomandibular joint neoplasms and pseudotumors. *Adv Anat Pathol* 2000;7:365-81.
2. Krishnapillai R, Angadi PV. A clinical, radiographic, and histologic review of 73 cases of ameloblastoma in an Indian population. *Quintessence Int* 2010;41:e90-100.
3. Halfpenny W, Verey A, Bardsley V. Myxoma of the mandibular condyle. A case report and review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2000;90:348-53.
4. Motamedi MH, Navi F, Eshkevari PS, Jafari SM, Shams MG, Taheri M, et al. Variable presentations of aneurysmal bone cysts of the jaws: 51 cases treated during a 30-year period. *J Oral Maxillofac Surg* 2008;66:2098-103.
5. Giddings NA, Kennedy TL, Knipe KL, Levine HL, Smith JD. Aneurysmal bone cyst of the mandible. *Arch Otolaryngol Head Neck Surg* 1989;115:865-70.
6. Motamedi MH, Stavropoulos MF. Large radiolucent lesion of the mandibular condyle. *J Oral Maxillofac Surg* 1997;55:1300-4.
7. Motamedi MH. Destructive aneurysmal bone cyst of the mandibular condyle: report of a case and review of the literature. *J Oral Maxillofac Surg* 2002;60:1357-61.

8. Park W, Nam W, Park HS, Kim HJ. Intraosseous lesion in mandibular condyle mimicking temporomandibular disorders: report of 3 cases. *J Orofac Pain* 2008;22:65-70.
9. Pelo S, Gasparini G, Boniello R, Moro A, Amoroso PF. Aneurysmal bone cyst located in the mandibular condyle. *Head Face Med* 2009;5:8.
10. Ettl T, Ständer K, Schwarz S, Reichert TE, Driemel O. Recurrent aneurysmal bone cyst of the mandibular condyle with soft tissue extension. *Int J Oral Maxillofac Surg* 2009;38:699-703.
11. Zlotogorski A, Buchner A, Kaffe I, Schwarts-Arsd D. Radiological features of central haemangioma of the jaws. *Dentomaxillofac Radiol* 2005;34:292-6.
12. Cheng NC, Lai DM, Hsie MH, Liao SL, Chen YB. Intraosseous hemangiomas of the facial bone. *Plast Reconstr Surg* 2006;117:2366-72.
13. Jadu FM, Pharoah MJ, Lee L, Baker GI, Allidina A. Central giant cell granuloma of the mandibular condyle: a case report and review of the literature. *Dentomaxillofac Radiol* 2011;40:60-4.
14. Tasanen A, von Konow L, Nordling. Central giant-cell lesion in the mandibular condyle. Report of a case. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1978;45:532-9.
15. Said-Al-Naief N, Fernandes R, Louis P, Bell W, Siegal GP. Desmoplastic fibroma of the jaw: a case report and review of literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2006;101:82-94.
16. Katagiri W, Nakazawa M, Kishino M. Benign fibrous histiocytoma in the condylar process of the mandible: case report. *Br J Oral Maxillofac Surg* 2008;46:e1-2.
17. Macan D, Cabov T, Uglesić V, Manojlović S, Kaleb S, Spicek J. Chondromyxoid fibroma of the mandible. *Br J Oral Maxillofac Surg* 2003;41:261-3.
18. Martínez-Mata G, Mosqueda-Taylor A, Carlos-Bregni R, de Almeida OP, Contreras-Vidaurre E, Vargas PA, et al. Odontogenic myxoma: clinico-pathological, immunohistochemical and ultrastructural findings of a multicentric series. *Oral Oncol* 2008;44:601-7.
19. Whear NM. Condylar hemangioma-a case report and review of the literature. *Br J Oral Maxillofac Surg* 1991;29:44-7.
20. MacLennan WD. Haemangioma of the mandibular condylar process. *Br Dent J* 1958;105:93.
21. Uotila E, Westerholm N. Hemangioma of the temporomandibular joint. *Odontol Tidskr* 1966;74:202-6.
22. Schwenzer N, Schwenzer N. A bone hemangioma in the articular process of the mandible. *ZWR* 1975;84:72-4.
23. DelBalso AM, Banyas JB, Wild LM. Hemangioma of the mandibular condyle and ramus. *AJNR Am J Neuroradiol* 1994;9:1703-5.
24. Alves S, Junqueira JL, de Oliveira EM, Pieri SS, de Magalhães MH, Dos Santos-Pinto D Jr, et al. Condylar hemangioma: report of a case and review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2006;102:e23-7.
25. Abramowicz S, Marshall CJ, Dolwick MF, Cohen D. Vascular malformation of the temporomandibular joint: report of a case and review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2007;103:203-6.

Reprint requests:

Yuk-Kwan Chen, DDS, MS
 Department of Oral Pathology
 Kaohsiung Medical University
 Chung-Ho Memorial Hospital
 100, Tz-you 1st Rd.
 Kaohsiung
 Taiwan
 yukkwa@kmu.edu.tw