

large incisions can complicate reelevation of the scalp for future craniotomy/cranioplasty or free tissue transfer.

Regional nonadjacent tissue transfer is also limited to very specific indications and locations. Occipital defects up to 10 cm × 8 cm can be closed by a pedicled trapezius flap. Smaller temporofrontal defects can be reconstructed using a temporoparietal fasciocutaneous flap. Larger defects with exposed neurocranial structures, alloplastic material, or other infection require free tissue transfer. However, these complicated patients are not optimal candidates for the more extensive and definitive reconstruction methods of distant pedicle flaps or microvascular free flaps, instead requiring a temporizing measure for wound closure.

The visor flap provides an innovative solution for closure of complicated scalp defects. It takes after Jadhav's previously reported bipedicled scalp flap used in the reconstruction of high-tension electric burns of calvarium, which provided coverage of large wounds involving necrotic scalp, calvarium, dura, and necrotic brain.<sup>4</sup> Due to the large size of the wounds, the donor sites necessitated split thickness skin grafts for coverage. The visor flap also takes into consideration Hartzell V-Y modification of a bipedicle perforator flap, which minimizes the respective limitations of the bipedicled flap as well as the V-Y advancement flap.<sup>5</sup>

The 7 patients who presented to us had comorbidities that did not favor prolonged anesthesia for microsurgical reconstruction. Hence, the bipedicled scalp flap with V-V modification provided a simple yet reliable alternative for wound closure. The bipedicled nature preserves a robust bidirectional flow through choke anastomosis.<sup>6</sup> The V-V modification enabled advancement of the scalp at the donor site over the convex skull, precluding the need for additional skin graft reconstruction for donor site coverage. All patients demonstrated complete healing by primary intention of the recipient site. Only 1 of 7 patients required healing by secondary intention at the donor site, since a residual 3 mm gap was left purposely to avoid excess skin tension upon closure—no additional split thickness skin grafts were required.

Unlike other local flaps, this wide, bipedicled flap with V-V modification leaves the donor site remote from the wounded recipient site, which avoids further compromise to the surrounding skin in the form of T-incisions. This technique preserves the blood supply to the flap while allowing redistribution of a large surface area of scalp tissue as well as V-V advancement along the convexity of the skull. It optimizes immediate closure of scalp tissue compromised by factors such as infection, radiation, or exposed hardware without burning bridges to more complex reconstructive options that may be utilized once the patient becomes a better surgical candidate.

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# A Giant Aneurysmal Bone Cyst in the Mandibular Condyle

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**Abstract:** Aneurysmal bone cyst (ABC) is a rare, rapidly expanding, locally destructive, and easily misdiagnosed lesion. An ABC of the condyle is rare. This report presents a 25-year-old female with a giant ABC in the left mandibular condyle. This patient was treated with surgical resection of the affected bone and immediate mandibular reconstruction using autologous bone. Follow-up to date showed no signs of recurrence. The clinical feature, imaging finding, pathogenesis, and treatment methods of ABCs are discussed.

**Key Words:** Aneurysmal bone cyst, mandibular condyle, reconstruction

A neurysmal bone cyst (ABC) is a benign, osteolytic, expansive, and hemorrhagic lesion of bone, which predominantly occurred in long bones metaphysis like the femur, the tibia, or the spine. The World Health Organization defines ABCs as “a destructive, expansile, benign neoplasm of bone composed of multiloculated blood-filled cystic spaces.”<sup>1</sup>

Its occurrence in the jaw is uncommon. Only 15 patients originating in the condyle have been reported in the English literature. Aneurysmal bone cysts usually affect patients under 20 years of age with no sex predilection.<sup>2</sup> The etiology and pathogenesis of ABCs is still unclear. The clinical signs and symptoms of these lesions are nonspecific, sometimes making diagnosis difficult. Diagnosis should base on clinical presentation, imaging, and histopathological examinations. This case presents a rare giant ABC of the left mandibular condyle, including the treatment and postoperative follow-up. The study was performed in compliance with the relevant policies of Institutional Review Board of Peking University Hospital of Stomatology.

## CLINICAL REPORT

In July 2014, a 25-year-old female patient presented with history of left preauricular region swelling associated with pain and limited mouth opening, which had progressively aggravated over the past 2 months. She also complained of pain on chewing and about a week ago she felt numb in this region. There was no history of trauma. Clinical examination revealed a 4 cm × 4 cm swelling in the left preauricular region with tenderness and relatively firm texture. The patient's interincisal opening was 25 mm. The range of motion in the left temporomandibular joint (TMJ) decreased, and there was no cracking.

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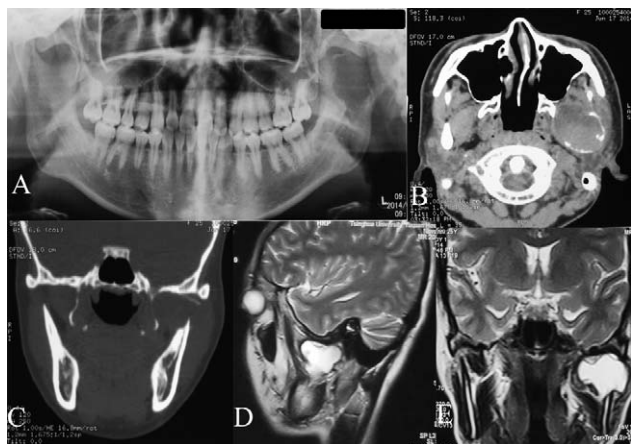
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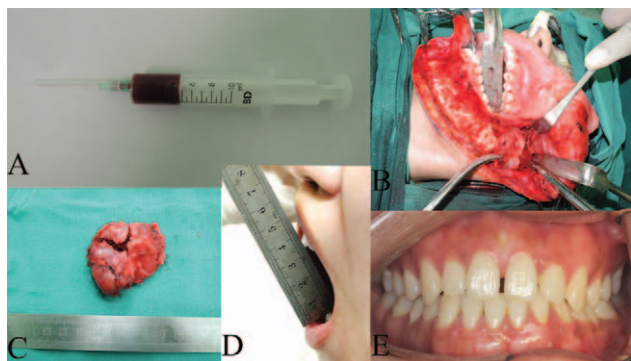
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**FIGURE 1.** Radiograph preoperatively. (A) Panoramic radiograph shows a large radiolucent lesion of the left condyle. Computed tomography scan shows expansion of the left condyle, with a bony rim. (B) Axial view. (C) Coronal view. Magnetic resonance imaging shows a lesion of the left condylar head with septa and fluid level. (D) Sagittal view. (E) Coronal view.

Panoramic radiographs showed a large expansile radiolucent lesion of the left condyle and mandibular ramus with unclear boundaries (Fig. 1A). Computed tomography (CT) showed a soft tissue mass replaced the condyle and the lesion appeared well circumscribed, with a thin cortical rim surrounding it (Fig. 1B and C). It extended to the left infratemporal space and masseteric space. But the glenoid fossa was intact. The primary impression is a malignant tumor. However, the following examination changed the impression. Magnetic resonance imaging (MRI) on T2-weighted images shows high-signal intensity demonstrating fluid/fluid levels within the condyle (Fig. 1D and E). Aspiration was performed and present incoagulable blood (Fig. 2A). Based on these findings, the preoperative diagnosis strongly suggested ABC.

On August 19, the patient received surgery under general anesthesia, which included excision of the lesion in the left condyle and partial mandibular ramus, reconstruction of the left condyle. Extraoral incision was made from the left submandibular area to the middle of the lower lip. The coronoid process and condyle were exposed. The lesion and a portion of ramus were resected (Fig. 2B and C). Condylar reconstruction was performed with grafting of the coronoid process and segment of the ascending ramus simultaneously. Intermaxillary fixation was performed postoperatively for 4 weeks, and the patient’s course was uneventful.



**FIGURE 2.** (A) Aspiration shows incoagulable blood. (B) Resection of the lesion during operation. (C) Resected specimen. Patient after the surgery. (D) Occlusion image. (E) Opening image.

The histopathological finding was ABC. Macroscopically, the lesion was surrounded by bony cortex with clear boundary, consisting of blood-filled cyst-like spaces of variable sizes. Microscopically, these spaces are lack of epithelial or endothelial lining, separated by fibrous tissue septa containing fibroblast, multinucleated giant cell, and osteoid tissue. Hemorrhage foci, hemosiderin deposition, phagocytic cells, and inflammatory cells can be observed.

On follow-up, clinical and radiographic examinations were taken after 1 month, 5 months, and 1 year and 7 months respectively; there was no evidence of recurrence (Fig. 2D). The patient revealed normal occlusion and the interincisal opening was 35 mm (Fig. 2E).

## DISCUSSION

Aneurysmal bone cysts were first reported by Jaffe and Lichtenstein in 1942. In 1958, a clinical case of an ABC of the jaw was reported by Bernier and Bhaskar. Aneurysmal bone cyst is an expansive, osteolytic bone lesion, which can expand rapidly and sometimes can be aggressive.<sup>3,4</sup> The characteristics of ABC histologically are the replacement of bone by fibro-osseous tissue containing blood-filled sinusoidal or cavernous spaces. The lesion generally affected young people, the peak incidence in the second decade of life and median age is 13, it was also significantly more common in the mandible than the maxilla.<sup>5</sup> In the English literature, only 15 cases of ABC located in the condyle that have been reported, the patients’ age is under 20-year old except one, female is predominant, only 3 patients have definitely trauma history. The main symptoms are swelling, pain, and mouth-opening limitation. Most patients have been treated with resection and reconstruction with no recurrence (Table 1).

The pathogenesis of ABC is controversial. Trauma may be the triggers that lead to the development of the ABC. In a research, about 50% to 70% patients can recall a history of trauma preceding the development, suggesting that trauma may be an initiating factor in its development.<sup>6</sup> But the relationship between trauma and ABCs has yet to be ascertained. In most clinical reports, the patients have no history of trauma. In the present patient, the patient denied any history of trauma and parafunctional habits.

Some theories have been put forward to try to explain the pathogenesis of ABC. One of the most widely accepted theories is that local haemodynamic alteration and arteriovenous malformations can increase venous pressures and expand the vascular bed, then leading to bone resorption, connective tissue replacement, and osteoid formation.<sup>7</sup> Whereas some other researchers deem that it can occur secondary to a preexisting lesion. Kershnik and Batsakis<sup>8</sup> reported that ABCs are presumably reactive lesions of bone and exist in 2 clinicopathological forms: either as a primary lesion (which may be vascular, solid, or mixed) or as a secondary lesion arising in other osseous conditions. Martineez and Sissons<sup>9</sup> reviewed 123 patients and found that in almost 30% of the patients, ABC existed in association with some other type of bone lesion, which is presumed to be antecedent to it. The most common associations were with solitary or unicameral bone cyst, and with osteoclastoma. Other associated lesions included osteosarcoma, nonosteogenic fibroma, osteoblastoma, hemangioendothelioma, and hemangioma of bone.<sup>10</sup>

Recently, cytogenetic studies indicate a neoplastic basis in at least some ABCs. Panoutsakopoulos et al<sup>11</sup> found that chromosomal translocation t(16;17)(q22;p13) contributed to the development of ABC. These observations were later confirmed by Dal Cin et al.<sup>12</sup> Oliveira et al<sup>13</sup> have shown that the chromosomal translocation t(16;17)(q22;p13) fuses the promoter region of the osteoblast cadherin 11 gene (CDH11) on chromosome 16q22 to

**TABLE 1.** Fifteen Patients of Aneurysmal Bone Cyst Located in the Mandibular Condyle

Patient	Age	Gender	Trauma History	Location	Symptoms	Surgery	Recurrence	Follow-Up
1993	14	M	N/A	Right	Swelling, pain, tenderness, opening limitation, anesthesia of the inferior alveolar nerve	Resection and reconstruction	No	2 years and 8 months
1997	11	F	Yes	Right	Swelling, pain, tenderness	Curettage	No	1 year
2000	12	F	Yes	Left	Swelling, pain, clicking, opening limitation	Resection and reconstruction	No	5 years
2002	18	M	No	Right	Swelling, tenderness, pain	1. Curettage 2. Resection 3. Resection	Yes Yes No	several months several months 5 years
2004	16	M	No	Left	Swelling, opening limitation	Resection and reconstruction	No	2 years and 8 months
2008	17	F	No	Right	Swelling, lock and clicking, tenderness	Curettage	No	7 years
2009	10	M	No	Right	Swelling	Resection	No	3 years
2009	17	F	No	Right	Swelling, pain, tenderness, opening limitation	1. Resection 2. Resection and reconstruction	Yes No	6 months 8 months
2011	17	F	No	Right	Swelling, pain, opening limitation	Resection	No	1 year
2012	37	F	No	Right	Swelling, facial paralysis	Resection and reconstruction	No	5 years
2012	18	F	No	Left	Swelling, pain, opening limitation	Resection	N/A	N/A
2013	19	F	No	Right	Swelling, opening limitation	Resection and reconstruction	No	6 months
2015	16	F	No	Left	Swelling, pain, opening limitation	Resection and reconstruction	No	7 months
2015	17	M	Yes	Right	Swelling, motion limitation	Sagittal split ramus osteotomy	No	3 years
2015	18	F	No	Right	Swelling, tenderness, opening limitation	Resection and reconstruction	No	1 year

1, first surgery; 2, second surgery; 3, third surgery; F, female; M, male; N/A, not available.

the entire coding sequence of the ubiquitin protease (UBP) USP6 gene (also known as Tre2 oncogene) on chromosome 17p13, suggesting that the pathogenesis of most primary ABCs involves up-regulation of USP6 transcription driven by the highly active CDH11 promoter. They also found rearrangements of CDH11 and/or USP6 in almost 70% of the patients in a subsequent study of 52 primary ABCs, but negative in the secondary ABCs. Kenney et al<sup>14</sup> reported a novel translocation of t(X; 9) (q26; q32) in a solid variant of ABC.

Clinical presentation of ABCs varies strongly. The majority of lesions presented with a nontender swelling that gradually expands, painlessly or painfully. The mandibular condyle or ramus lesions presented with various degrees of limited mouth opening.<sup>15</sup> In a review of ABC located in mandibular condyle, most of patients manifested pain and/or tenderness upon palpation. Temporomandibular joint disorder symptoms like limited function, clicks, and malocclusion could also be observed. No parasthesia was reported in relation to condylar lesion per se, although self-limited postoperative neuropathy occasionally occurred.<sup>16</sup> Aneurysmal bone cysts usually present a slow progressive growth until cortical plates are eroded at any point and then show a rapid growth.<sup>17</sup> Due to the nonspecificity of its clinical presentation, it is important to differentiate it from other pathologies that occur in the maxillofacial region. These include peripheral and central giant cell reparative granulomas, traumatic bone cyst, brown tumor of hyperparathyroidism, myxoma, fibrous dysplasia, desmoplastic fibroma, fibrous histiocytoma, hemangioma, osteogenic sarcoma, globulomaxillary cyst, hemangioendothelioma, and hemangiopericytoma.<sup>6</sup>

Compared with the conventional radiography, CT is far superior in defining the extent of the lesion, especially in the mediolateral dimension. Intralesional calcifications may be present. Magnetic resonance imaging is more accurate than CT in depicting fluid levels or the presence of septa within the lesion.<sup>9</sup> To get the definite diagnosis of ABC is difficult due to the clinical and radiographic

similarities with some other pathologies. The radiologic findings of the aneurysmal bone cyst are suggestive but not diagnostic. Plain radiography and CT may result in misdiagnosis. Therefore, the fluid levels present in MRI are essential to diagnose ABC. In the present patient, CT only showed expanded, cystic lesion of the mandibular condyle. The fluid levels and the septa revealed by MRI image help to affirm the diagnosis.

A variety of treatments have been recommended for ABC, including arterial embolization, injection of sclerotherapy, excision or curettage, cryotherapy, radionuclide ablation, radiotherapy, or a combination of these modalities. The management of ABC depended on the age of the patient, extent, aggressiveness, size and localization of the lesions, ranging from simple curettage to extensive resection. According to Sun et al,<sup>18</sup> most ABCs were combined with another pathologic entity, such as ossifying fibroma, central giant cell granuloma, or benign osteoblastoma. This might indicate that curettage is not an appropriate treatment of ABCs. In a review of the literature of 1256 aneurysmal bone cysts,<sup>19</sup> the authors suggested that inactive lesions can heal with biopsy or curettage alone. In active or aggressive lesions, resection offers a satisfactory theoretical solution. When the extent of lesion is large and the resection will result in damage of TMJ, reconstruction of the TMJ is essential. Reconstruction of the damaged TMJ is dependent on the cause of damage and the patient's age. Of the 15 patients reviewed, 3 patients had been treated with curettage, 12 with resection. In the 3 patients with curettage, 2 recurred. Those patients with resection had only 1 recurrence, except that 1 with no follow-up information. Nine patients reconstructed the TMJ using different methods (9/12), 5 TMJs were reconstructed with costochondral graft (5/9), 1 with costochondral graft and particulate cancellous bone marrow of the iliac crest (1/9), 2 with artificial joint (2/9), and another one with sagittal split ramus osteotomy to reconstruct the condyle (1/9). Autogenous grafts for condylar reconstruction are the most widely used grafts for condylar reconstruction.<sup>20</sup> In childhood the current preference is for autogenous reconstruction that can potentially



“grow” with the child. While in the adult, considering the limitation of autogenous bone supply and morbidity of the donor site, the alloplastic reconstruction is another choice that gives a more stable long-term result and facilitates early mobilization.<sup>21</sup>

In the present patient, it is difficult to expose and remove the lesion completely with conventional preauricular approach because of its large size. The surgical approach was determined by trans-mandibular approach to explore the lesion thoroughly. Then the condylar reconstruction was performed with graft of the neighboring coronal process and anterior part of ascending ramus to recover the height of the condyle. Postoperatively, intermaxillary fixation was performed postoperatively for 4 weeks for a stable occlusion. Follow-up to date showed no signs of recurrence and the patient had a good occlusion relationship and mandibular movement. Anyway, the long-term follow-up is still necessary because of the possibility of recurrence and bone resorption.

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## Magnet-Retained Orbital Prosthesis Using a Dental Implant

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**Abstract:** The loss of an eye and the associated facial disharmony has major physical, psychological, and social consequences for patients undergoing orbital exenteration. A magnet-retained prosthesis with an implant has various advantages over both adhesive and spectacle-retained prostheses for reconstruction of the exenterated orbit.

The author demonstrates one representative patient with our orbital reconstruction patients with magnetic implants, which will be applied to various maxillofacial prosthesis strategies in the near future.

**Key Words:** Magnet-retained prosthesis, orbital exenteration, orbital implant

The use of cranial implants for prosthetic reconstruction in the facial region has been developed recently with the use of dental implant as a kind of innovative works.<sup>1–4</sup> For avoiding several retention problems in the orbital prosthetic reconstruction, this clinical report presents another way with magnetic abutment.

## CLINICAL REPORT

A 75-year-old man was diagnosed with squamous cell carcinoma of the right maxilla. He underwent mass excision, partial maxillectomy, right supraomohyoid neck dissection, and reconstruction with radial forearm free flap. After 8 months, further hemi-maxillectomy, orbital floor excision and exenteration were performed due to

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