Aneurysmal bone cysts (ABCs) are rare benign tumors with a prevalence of 0.14 cases per 100,000 people.11 A majority of cases arise during adolescence, and there is a female predominance. This lesion accounts for 1.4% of all primary bone tumors. Aneurysmal bone cysts occur mainly in the long bones, with spinal involvement in 10–30% of cases. Cervical spine ABCs account for about one-third of spinal ABCs, and atlas involvement occurs in 1% of cases. Resection of ABCs at the atlas is difficult because of the location and the lack of proper instrumentation for reconstruction of C-1. The authors present a case of an ABC at C-1 in a child who underwent resection of the lesion and reconstruction of the lateral mass with a titanium mesh cage. (DOI: 10.3171/2008.10.SPI08403)

KEY WORDS • aneurysmal bone cyst • atlas • lateral mass • titanium cage

Case Report

History and Examination. This 12-year-old girl presented to our institution with a 2-month history of progressively worsening neck pain. The pain was severe enough that she was unable to sit upright without supporting her head with her hand. She had no history of trauma. On examination, she had a normal neurological examination, including normal motor and sensory examinations. Computed tomography scanning of her neck revealed an expansile, lytic lesion, measuring 4.4 × 3 cm and centered in the right lateral mass of C-1 (Fig. 1). The lesion was isodense to bone on T1-weighted images and hyperdense on T2-weighted images with heterogeneous enhancement. Radiographic findings were suggestive of an ABC or giant cell tumor. A CT-guided biopsy of the lesion yielded blood, histiocytes, and multinucleated giant cells consistent with the diagnosis of an ABC. Preoperative angiography revealed collateral filling from the tumor feeding vessels to the anterior spinal artery, making serial embolization as the primary treatment impossible. Given the extreme severity of the patient’s symptoms and the results of biopsy sampling, the patient and her parents elected for resection of the lesion and immediate reconstruction of the lateral mass with a titanium mesh cage.

Operative Technique. The patient underwent preoperative angiography, which revealed a patent right VA displaced and narrowed by the tumor. The blood supply to the tumor came from a posterior ascending artery and an ascending pharyngeal artery; however, because these 2 arteries had extensive collateral vessels to the high cervical spinal artery, cavernous internal carotid artery, and VA, no embolization could be performed. The right VA was occluded endovascularly from the intradural portion down to the C-2 segment to minimize intraoperative blood loss and avoid the significant risk of intraoperative VA injury.
The patient was placed in a prone position with the Mayfield headset. A midline incision extending from the occiput to C-4 was made and soft tissue was dissected free from the bone in a subperiosteal fashion to expose the occiput as well as the spinous processes, facets, lamina, and lateral masses of Cl–4. A midline occipital plate was placed, and 3 screws with bilateral cortical purchase were used to fixate the occipital plate. A lateral mass polyaxial screw was then placed on the left side of C-1, and a pedicle screw was placed on the right side of C-2. Because the right VA had already been occluded endovascularly, no pedicle screw was placed on the left side so that potential injury to the left VA would be avoided. Bilateral C-3 and C-4 lateral mass screws were placed using the Magrel technique.

A rib graft was then harvested from the right side from the T-7 rib. An iliac crest graft was harvested from the left iliac crest. Tumor resection began after obtaining the bone graft. The posterior arch of C-1 was identified and dissected free from soft tissue. It was removed using Kerrison rongeurs. The tumor was readily identified as a destructive mass involving the right side of the C-1 posterior arch. The coiled right VA was then mobilized. It was ligated by hemoclips and transected through the coils in the VA. The right C-1 and C-2 nerve roots were then identified and ligated. The right lateral mass of C-1, which was destroyed by the tumor, was removed with a pituitary rongeur and curette from the occipital condyle through the superior articular pillar of C-2. The tumor was followed anteriorly toward the retropharyngeal space and removed using suction and a pituitary rongeur; the tumor capsule was dissected free with curettes and Penfield dissectors. Gross-total resection of the tumor and capsule was achieved. Hydrogen peroxide was applied to the resection cavity for hemostasis and local tumor control.

To restore the load bearing property of the C-1 lateral mass, a titanium cage was used to reconstruct the C-1 lateral mass. A 16-mm pyramesh cage was contoured to fit the occipital condyle and the superior surface of the articular pillar of C-2 lateral mass and filled with the autograft material from the iliac crest. The occipital condyle and the C-2 lateral mass were decorticated before the cage was placed. A screw was placed through the cage to secure it to the occipital condyle (Figs. 2 and 3).

Two 3.5-mm rods were then placed over the occipital plate and the lateral mass screws, and the rods were secured in place with set screws. Further compression of the rod was applied across the titanium cage. The rib graft was then cut in half; 1 piece was placed on the right side from the occipital condyle to the C-3 lateral mass and stabilized with microfracture screws, and the other half was placed on the left and secured with a C-2 sublaminar wire. Additional stability was provided with a cross-link. The wound was irrigated extensively and closed in layers. The estimated intraoperative blood loss was 900 ml.

**Postoperative Course.** The postoperative course was uneventful. The patient remained neurologically intact and was transferred out of the intensive care unit after 2 days. Postoperative CT scans and MR images revealed gross-total resection of the tumor. The patient underwent in-patient physical therapy and was discharged on postoperative Day 8 without any complication. She was maintained in a cervical collar for 12 weeks. At her 6-month postoperative visit, CT scanning showed no recurrence of tumor. There were signs of fusion with bony trabeculae extending from the occipital condyle through the titanium condyle.
Reconstruction of C-1 lateral mass with titanium cage

cage to the C-2 articular pillar, without evidence of hardware failure (Fig. 2). Despite the extensive instrumentation, the patient had a good range of motion; she had no significant pain and no neurological deficits. Currently, the patient has a neck Visual Analog Scale score of 2 and Short Form-36 Physical Component score of 42.

Discussion

Aneurysmal bone cysts are rare benign tumors that primarily affect adolescents.²,¹¹,¹⁶ Spinal ABCs account for 10–30% of ABCs.²,¹⁵,¹⁶ Pain is the most frequent presenting symptom in patients with spinal ABCs, and neurological deficits are uncommon.²,¹⁵,¹⁶

Traditionally, ABCs are treated with resection. En bloc excision of the tumor is associated with less blood loss and may be associated with lower recurrence rates; however, resection is quite difficult in the high cervical spine. Intralesional curettage and resection is the most commonly used surgical strategy. Complete excision with intralesional curettage is associated with the extremely low recurrence rate, whereas incomplete excision has been reported in recent studies to carry a 5–10% recurrence rate.²,¹⁶ In the past, incomplete excision was more likely to be caused by significant intraoperative blood loss and lack of instrumentation for surgical stabilization after radical resection of bone elements. However, modern techniques such as preoperative embolization of feeding vessels, and the availability of spinal instrumentation allows for a more radical excision.

Selective arterial embolization is an alternative to surgical excision and has been used extensively in skeletal ABCs with a success rate similar to curettage.² The published experience with spinal ABCs treated with SAE is limited due to the tumor’s rarity and the relatively short history of SAE.² Treatment of ABCs with SAE often require multiple sessions, and improvements are usually seen over the course of several months.¹⁰,¹⁴ After successful embolization, the decrease in the size of the soft-tissue mass and ossification of the lesion can be seen on CT.¹⁰,¹⁴ In our patient, unfavorable spinal cord vascular supply from the tumor feeding vessels, as well as the extreme severity of symptoms, made embolization as a primary treatment and adjuvant preoperative treatment impossible.

Radiation therapy has also been used in the treatment of spinal ABCs. Historically, RT led to local control of ABCs at a rate that is comparable to surgery in several small series.⁵ Limited data suggest that local control of spinal ABCs can be achieved, although recurrence is not uncommon.³ In addition, RT is associated with the potential risk of secondary cancer, and therefore, RT has

Fig. 2. Plain sagittal (A) and coronal (B) radiographs obtained 6 months after the operation show no evidence of hardware failure or spine malalignment. C: Computed tomography scan obtained 6 months postoperatively demonstrates signs of fusion with bony trabeculae extending from the occipital condyle through the titanium mesh cage. The rib graft is also shown to be solidly fused (D).
mostly been used in cases of surgically inaccessible lesions, or as an adjuvant therapy.²,³,⁵

Aneurysmal bone cysts of the atlas are extremely rare and estimated to comprise 1% of all spinal ABCs.⁴ Review of the literature revealed 4 case reports of ABCs of the atlas.¹⁷,¹⁴,¹⁷ Other authors reported their experience with the treatment of atlas ABCs in surgical series of CCJ tumors.⁶,¹² In most cases, the ABC of atlas was located in the lateral mass.⁷,¹²,¹⁴

Treatment for ABCs of the atlas is difficult and associated with significant complications due to location, proximity of the VA, and the lack of reconstruction tools after resection. In addition to resection, SAE and percutaneous intralesional injection of calcitonin and methylprednisolone have both been tried successfully in the treatment of ABCs of the atlas.⁷,¹⁴,¹⁷ However, both treatments require multiple sessions of therapy that take effect over the course of several months.⁷,¹⁴ Patients who undergo this type of treatment will often will need to wear a cervical collar during the course of therapy.⁷ Selective arterial embolization in the high cervical region is also associated with the risk of spinal cord infarct.²

Resection of ABCs in the atlas is still the most common treatment for these lesions reported in the literature. However, surgery in the CCJ is difficult due to the proximity of the brainstem and spinal cord. The VA often must be mobilized and transposed during the exposure.⁶,¹² The anatomy of the CCJ also presents unique challenges to reconstruction of the region. The atlas is unique in that it lacks a vertebral body. It has 2 large lateral masses that articulate with the occipital condyle and the superior surface of the articular pillar of axis.⁵ The 2 lateral masses of atlas therefore transmit the load from the occipital condyle to the C-2 vertebra. In the past, reconstruction of the lateral mass of the atlas involved the placement of iliac crest graft in the region of the lateral mass after tumor excision.¹⁶ This may be supplemented with addition rib graft and lamina wire for a posterior fusion.¹,⁶,¹²

We chose to resect the lesion and then immediately reconstruct the C-1 lateral mass with a titanium mesh cage in our patient. Her symptoms were quite severe and limited her activity of daily living. Moreover, the relatively large size of the lesion and extensive destruction of the bone also carried the potential danger of pathological fractures and neurological compromise during the treatment period. Selective arterial embolization was not a viable treatment option in this case because of the extensive collaterals of the 2 feeding blood vessels. Intralesional injections of calcitonin and steroids often take several months to take effect. Radiation therapy also requires multiple sessions and may be associated with the risk of secondary cancer. Moreover, RT for this lesion may expose the brainstem region to radiation due to the location of the tumor. In view of all of these factors, we chose resection was chosen for treatment of this tumor.

We used a posterior approach to resecting this tumor. Although most of the lesion was in the lateral mass, some was in the posterior arch region, and a laminectomy was required to access this part of the tumor. Through a posterior approach, we performed the resection, reconstructed the lateral mass, and achieved posterior instrument placement and fusion from the occiput to C-4 in a 1-stage procedure. Because complete resection of the tumor including the tumor capsule was achieved, no adjuvant therapy was necessary in our patient.

To minimize blood loss and risk of injury during resection, the right VA was occluded endovascularly before the operation; the procedure was simplified by the preoperative VA sacrifice but it was not required. If preoperative angiography or test occlusion had demonstrated that the right VA could not be sacrificed, we could have skeletonized and transposed it from the transverse foramen at C-1 and gently retracted it as required for exposure for the resection. The risk of VA injury and additional blood loss would of course be higher with this technique.

We reconstructed the lateral mass with a small titanium mesh cage. There has been extensive experience of using titanium cage to reconstruct the cervical vertebral

Fig. 3. Artist’s representation of the tumor (A) and reconstruction of the C-1 lateral mass with a titanium cage after resection (B). The load bearing property of the C-1 lateral mass is restored by the titanium cage.
Reconstruction of C-1 lateral mass with titanium cage

Biomechanical studies have shown that reconstruction with a titanium cage after corpectomy is as biomechanically sound as other structural graft materials. We were able to contour a titanium pyramesh cage filled with autograft to fit snugly between the occipital condyle and the superior articular surface of the C-2 articular pillar. This is the mechanism by which the weight of the head is transferred to C-2 and is thus similar to placing a load bearing graft or cage elsewhere in the anterior column. We used a small screw to further secure the cage to the occipital condyle. This effectively reconstructs the right lateral mass of C-1 and restores the load bearing properties of the atlas. Further stability is provided by instrumentation and fusion from the occiput to C-4. To our knowledge, ours is the first report of reconstruction of a lateral mass with a titanium cage.

Conclusions

We presented a case of ABC of the atlas. These are rare lesions of the cervical spine but treating them is often challenging. Although complete resection and reconstruction is the treatment of choice, it is often difficult. We performed a complete resection of an ABC of the C-1 lateral mass in our patient via a posterior approach. We also described our technique for reconstructing the lateral mass of C-1 with a titanium mesh cage. This reconstruction technique will also be applicable to other resection surgeries involving the CCJ.

Disclaimer

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

References


Address correspondence to: Christopher P. Ames, M.D., Department of Neurological Surgery, University of California San Francisco Spine Center, University of California, San Francisco, California 94143. email: amesc@neurosurg.ucsf.edu.