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A Case of C 3 Aneurysmal Bone Cyst Managed by Staged Surgery

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Abstract

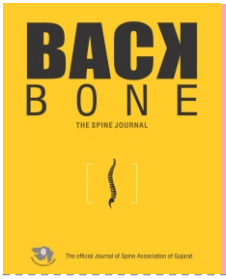
Aneurysmal bone cysts are often mistaken as malignant tumors like lesions or another benign bony lesion because of their proliferative component. Treatment of spinal aneurysmal bone cyst is challenging because of its proximity to the spinal cord, unique pathology, and complex growth characteristics. The treatment options are curettage and bone grafting, irradiation, embolization, intralesional injection of calcitonin, and steroid. We present a case of cervical aneurysmal bone cysts operated in a staged procedure: arterial embolization followed by surgical resection and stabilization.

Keywords: Aneurysmal bone cyst, Arterial embolization, Bone grafting.

Introduction

Aneurysmal bone cysts are benign, highly vascular tumors, locally aggressive, constituting around 1 % of all bony tumors [1, 2]. The lesion occurs primarily in the first two decades of life with female predominance [3]. Sankerin et al. described the solid variant of an aneurysmal bone cyst apart from a conventional cyst [4]. Lumbar is the least common presentation, followed by cervical and thoracic in vertebral bone.

Pain is the most common symptom, which is localized and often present at night. The duration of symptoms at presentation is usually <6 months. Neurological symptoms are uncommon but in severe form; paraplegia, cord compression and cauda equina syndromes are often present. It is often mistaken as a malignant tumour or another bony lytic lesion, and treatment modalities depend excessively on diagnostic parameters. On histology, microcellular proliferation is a predominant component, and typical mitotic figures can be found in reasonable numbers in more cellular areas. A variety of treatment options are available depending on the extent and location of the tumor. We present a case of a 35-year-old male suffering from neck pain for the past 2 years, diagnosed with an aneurysmal bone cyst of the cervical vertebra and underwent surgical resection and posterior stabilization.



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Case Report

A 35-year-old male presented in the outpatient department with complaints of neck pain. It was nagging in nature and was radiating over his left shoulder. The pain was present during rest and caused disturbance of sleep. It increased over the past 2 weeks and hampered his day to day activities. History was unremarkable. On examination, gait was unaltered, and neck movements were restricted. Neurology and circulation in both upper and lower limbs were normal. Plain radiographs showed an expansile lytic lesion in the spinous process of the third cervical vertebra (Fig. 1). Magnetic resonance imaging (MRI) imaging revealed fluid-filled (low signal on T1 and high signal on T2) multilobulated expansile cystic lesion in the third cervical vertebra more on the left side with well-defined margins. It encroached the vertebral artery foramen on the left side.

Computed tomography (CT) scan showed expansile, irregular bony cortical destruction involving the spinous process, lamina, lateral mass and vertebral artery foramen more on the left side. MR angiography showed the tumour receiving blood supply from ascending cervical artery and deep cervical arteries on both sides(Fig. 2a-c). The patient was treated in a staged procedure. Initially, he underwent embolization of the feeding arteries (ascending cervical artery and deep cervical arteries on both sides), and N-Butyl cyano acrylic glue was injected percutaneously (Fig. 3a and b). Within 24 hours, he was taken for surgical excision. Under general anaesthesia, prone position, posterior midline incision, the tumor mass was identified. The tumor was carefully removed in piece meal (Fig. 4a and b). After decompression, posterior stabilization was done with pedicle screw fixation(Fig. 5). A drain was

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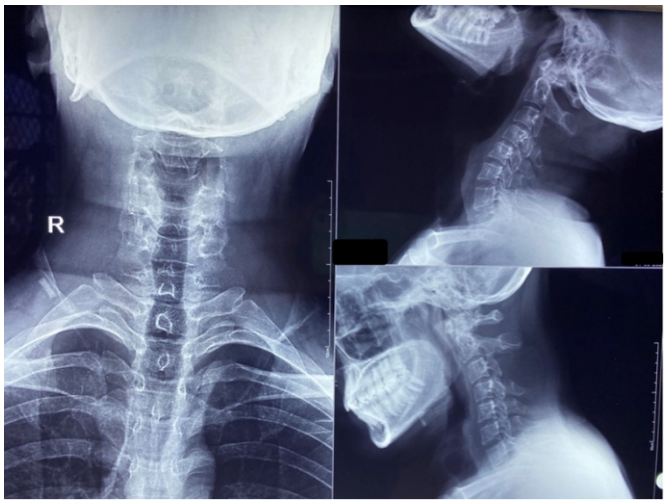

Figure 1: X- ray with expansile lytic lesion in the spinous process of C 3 vertebra.

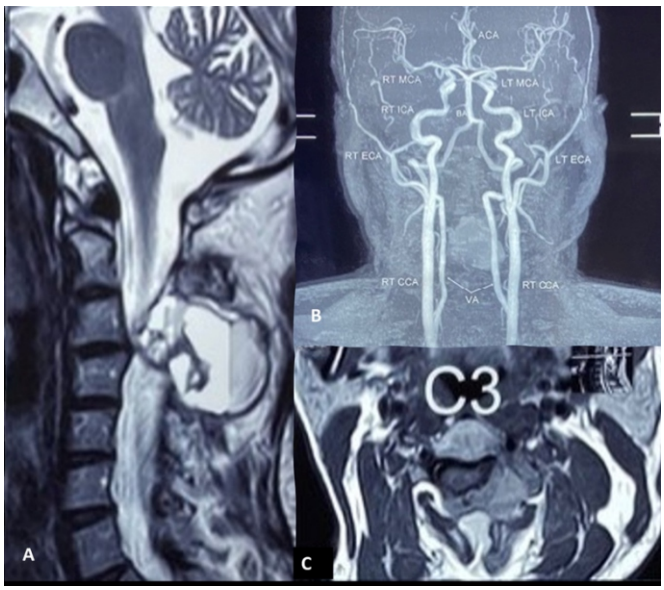

Figure 2: a. Magnetic resonance imaging sagittal view with a fluid-filled multilobulated expansile cystic lesion in the third cervical vertebra, b. MR angio showing tumor receiving blood supply from ascending cervical artery and deep cervical arteries on both sides, c. Magnetic resonance imaging axial view tumor enchoring vertebral artery on the left side.

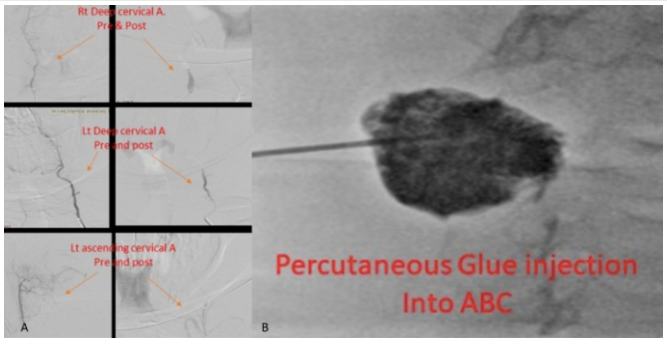

Figure 3: a. Embolization of the feeding arteries, b. Intralesional glue injection.

placed, and closure was done in layers. The tumor was pinkish red tissue grossly of 5 cm × 4 cm. Histopathological reports confirmed aneurysmal bone cyst with osteoclastic giant cells, spindle cells, blood vessels without atypical mitosis, and necrosis.

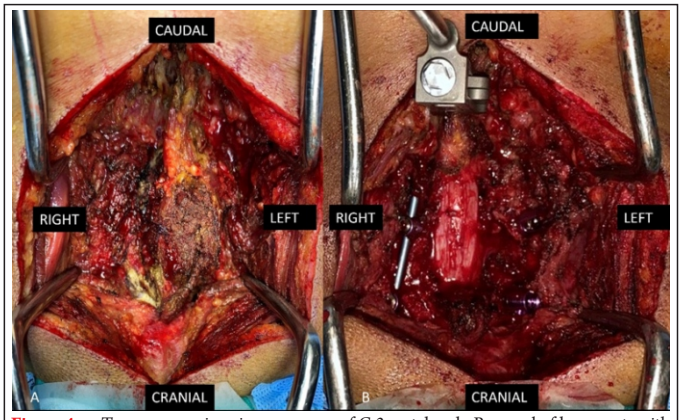

Figure 4: a. Tumour mass in spinous process of C 3 vertebra, b. Removal of bone cysts with posterior stabilisation of vertebra.

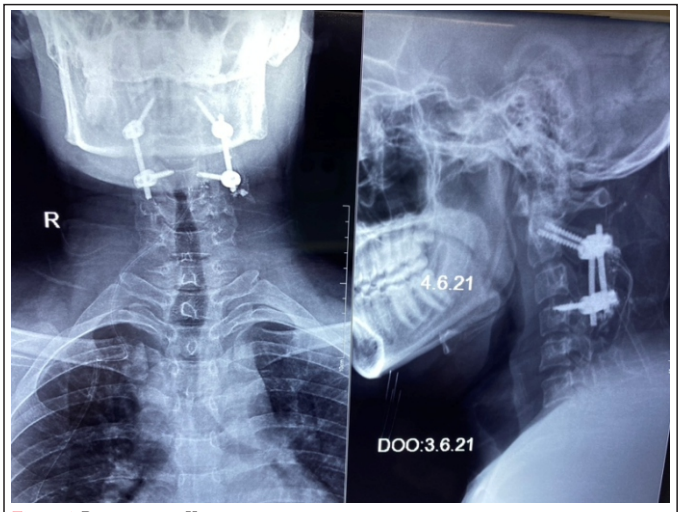

Figure 5: Post-operative X-ray.

Discussion

An aneurysmal bone cyst is a benign, highly vascular, locally aggressive tumour [2] with a hereditary component of disease [5]. Bertoni reported that the patient age distribution was 2–49 years (mean, 23 years), and the male/female ratio was 1:1.5 [6]. In our case, the patient was of 35 years, similar to Bertoni. The femur and tibia were the most commonly affected sites, and the spine was rarely involved. A spinal aneurysmal bone cyst is challenging because of its unique pathology, growth characteristics and complex neighbouring anatomy. Differential diagnosis of ABCs includes giant cell tumor, chondroblastoma, chondromyxoid fibroma, fibrosarcoma, telangiectatic osteosarcoma, fibrous dysplasia, simple bone cyst, osteoblastoma, and plasmacytoma [7].

Aneurysmal bone cysts originate in the posterior neural arch and expand unilaterally to produce an eccentric paravertebral lesion. Vertebral body involvement is rare, and as a result, body collapses are not often seen on X-rays. On the other hand, granular cell tumors usually arise from the vertebral body and spare the posterior elements [1]. In our case, plain radiographs showed an expansile lytic lesion in the spinous process of the third cervical vertebra. Capanna et al. confirmed dorsal

involvement in each of 22 examined spinal ABCs, with the asymmetrical involvement of the vertebral body in 12 cases and symmetrical involvement in two patients, and the posterior vertebral arch solely involved in eight of these cases [8].

Both CT scans and MRI are essential tools for planning for surgical treatment [1]. CT scan usually presents with multiloculated cystic lesion with multiple internal septations, pathological fracture or vertebral body collapse. Radiographic imaging hallmarks of Giant cell tumors include osteolysis and expansion without a marked sclerotic border [9]. In our case, similar expansile, irregular bony cortical destruction involving the spinous process, lamina, lateral mass and vertebral artery foramen with no sclerotic border were found.

MRI shows a thin, well-defined rim of low instability in the periphery and multiseptated lesion [10]. Multiple fluid levels were identified in the cysts, with the inferior portion showing a decrease of signal intensity, while the upper part showed an increased signal [3]. Murphy et al. reported that giant cell reparative granuloma, which is synonymous with the solid variant of aneurysmal bone cysts, has low to intermediate signal intensity in both T1- and T2-weighted MRI [11].

In our case, similar findings of a fluid-filled multilobulated expansile lytic lesion were found. In a spinal aneurysmal bone cyst, the cystic components can be completely absent. A solid benign bone tumour is depicted, making it impossible to differentiate these from other benign bone tumours, such as giant cell tumours.

Only histological analysis can definitively diagnose the type of tumor in these cases [12]. The lack or presence of anaplasia in histology studies argues against or support the diagnosis of a malignant tumor such as osteosarcoma or fibrosarcoma. The histological features of the aneurysmal bone cyst are characterized by Sanerkin et al. (1) florid fibroblastic or fibrohistiocytic proliferation without any cellular or nuclear pleomorphism, (2) areas rich in osteoclast-like giant cells, (3) osteoblastic differentiation with osteoid production, (4) aneurysmal sinusoids, and (5) occasional foci of degenerate calcifying fibromyxoid tissue [4]. On histology, microcellular proliferation is a predominant component, and typical mitotic figures can be found in reasonable numbers in more cellular areas. Accurate histological evaluation with a correlation of radiographic and MRI findings is imperative for definitive diagnosis [14]. In our case, there were osteoclastic giant cells, spindle cells, blood vessels without atypical mitosis and necrosis, which ruled out a malignant tumor.

There are multiple treatment modalities, but the treatment depends upon the following aspects, the tumour's proximity to the spinal cord, postoperative spinal stability, the necessity to remove the extensive lesion, its relative inaccessibility and intraoperative bleeding. The treatment options are curettage and bone grafting, irradiation, arterial embolization and intralesional injection of calcitonin and steroid [17]. Gladden et al. chose percutaneous intralesional injection of calcitonin combined with methylprednisolone acetate in the pediatric population [15]. They combined the proposed angiostatic and fibroblastic inhibitory effects of methylprednisolone with the presumed promotion of new bony trabeculae formation and osteoclastic inhibitory effects of calcitonin [15].

Radiotherapy is an effective treatment for aneurysmal bone cysts but is not suitable for primary treatment, as it carries the risk of post-radiation osteosarcoma, especially in children. Radiation therapy may lead to post-irradiation sarcomas, myelopathy, and deformity in patients with conventional aneurysmal bone cysts; it should be reserved for patients with inoperable lesions because of location or associated medical conditions aggressive recurrent disease [1, 12].

Complete resection remains the treatment of choice, particularly in patients with neurological deficits [15]. Operating in a staged manner, i.e., firstly embolizing the feeding artery and intralesional glue injection and removing the tumor in the second stage, gave excellent results. Intralesional glue injection will lead to less blood loss. Embolization should be performed before intralesional excision, especially in technically challenging sites (such as the cervical spine and sacrum) where there is a high risk of cord damage, pathological fracture, or nerve root or cord compression [1]. Subtotal excision may lead to the recurrence of the tumour up to 25% [16]. If local recurrence occurs, it is difficult to manage and result in significant impairment. Extensive tumour resection can also cause postoperative iatrogenic instability. If instability and deformity exist or the amount of bone removed leads to instability, then simultaneous reconstruction and instrumentation are recommended [7].

In our case, after embolization and intralesional glue injection, the tumor was excised, and the spinal instability was corrected by posterior fixation of the cervical vertebra. Postoperatively the patient was pain-free with normal neurology. On 1 year follow-up, the patient was pain-free, and MRI showed no recurrences.

Conclusion

The treatment modality of spinal aneurysmal cysts should be decided according to the tumour's proximity to the neurovascular structures. Performing the surgery in a staged manner, embolization of feeding arteries followed by intralesional percutaneous glue insertion and surgical excision of the tumour mass leads to less operative bleeding. After resection of the tumour, spinal instability, if present, should be corrected by posterior stabilization.

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Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the Journal. The patient understands that his name and initials will not be published, and due efforts will be made to conceal his identity, but anonymity cannot be guaranteed.

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