Case Report

Locally aggressive aneurysmal bone cyst of C4 vertebra treated by total en bloc excision and anterior plus posterior cervical instrumentation

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Abstract

We are presenting a case of cervical (C4) aneurysmal bone cyst in a 13-year-old girl, came to the outpatient department with neck pain and stiffness since 6 months and normal neurology. We did an en bloc excision of locally aggressive tumor through anterior plus posterior approach and stabilization by lateral mass screw fixation and anterior cervical instrumentation. Involvement of several adjacent cervical vertebrae by an aneurysmal bone cyst is rare, and conventional treatment with curettage and bone grafting is most likely to carry a high rate of recurrence and spinal instability. We recommend complete excision of the tumor and instrumentation in a single stage to avoid instability.

Key words: Aneurysmal bone cyst, anterior cervical stabilization and fusion, lateral mass screw fixation, total en bloc excision

INTRODUCTION

Aneurysmal bone cyst (ABC) is an expansile lytic lesions, comprising thin walled cystic cavities filled with blood. These lesions are more commonly seen in pediatric age group with a slight predominance in females. They comprise 15% of all kinds of primary spine tumors.[1] Although they can be located in all bone types, they are often found in flat bones, the pelvis, and the metaphysis of long bones. Approximately 10-30% of such cysts affect the spine, mostly the thoracic and the lumbar regions.[2-4,6]

Within the spine, the lesion spans the posterior elements, extend in an anterior manner to include the pedicles, the vertebral bodies, and the paraspinal areas. About 2% of ABCs are encountered in the cervical spine and generally involve the laminae, pedicles, spinous processes and occasionally extend to the facet joints and vertebral body.[3]

Total en bloc excision is usually associated with a good outcome but may be difficult to achieve in the cervical spine due to the close proximity of neurovascular structures and resultant instability. Fusion and stabilization with instrumentation have often been avoided in children due to concern over instability following future axial skeletal growth.

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CASE REPORT

A 13-year-old girl came to the outpatient department with the history of chronic neck pain and stiffness since last 6 months. The pain was limited to the cervical region, constant in nature, and increased since 15 days. There was no radiation of pain, no diurnal variation. There was no history of fever and weight loss.

During examinations, neck movements were found to be restricted in all planes by 50-60%. Local tenderness at upper and mid cervical region was there. Neurovascular examination was normal. Laboratory investigations included complete blood count, erythrocyte sedimentation rate, and general chemistry; all were within reference ranges.

The patient was first evaluated by a standard X-ray of the cervical spine, which displayed huge lytic lesion involving more of C4 and part of C3-C5 vertebrae with anterior translation of C4 over C5. Lesion extended more on the left side. There was collapse of left half of C4 body resulting in scoliosis. C3-C4 and C4-C5 disc space was reduced with loss of normal cervical lordosis. Soft tissue shadow in front of the lesion was normal [Figure 1].

Further evaluation with a computed tomography (CT) scan showed a large expansile multiseptated mass, containing fluid-fluid levels, involving C4 and part of C3 and C5 vertebrae. Lesion was involving whole C4 vertebral body, its posterior elements, and left lateral masses of C3 and C5 vertebrae [Figure 2].

Magnetic resonance imaging (MRI) of the cervical spine displayed a large expansile multilobulated lesion involving the whole body of C4, posterior elements mostly on the left side, and part of C3 and C5 lateral masses on left side. The lesion also had a large extraspinal component. The left vertebral artery was in a place surrounded by the tumor. There was no cord compression. The cysts were heterogeneous in appearance according to both T1- and T2-weighted images. The presence of multilobulated cysts containing fluid-fluid interfaces on T2 weighted images was characteristic and highly suggestive of the diagnosis [Figure 3].

Selective embolization of the tumor was done 24 h prior to surgery.

After the embolization, the tumor was exposed through a standard posterior midline incision. After dissection through fascial planes, it was found that the lesion had eroded the lamina of C4 and the left lateral masses of C3, C4, and C5 vertebrae. It had a multilobulated, sponge-like appearance, and an egg shell like thin rim of bone. After resection of the posterior paraspinal tumor and the involved bone (C4 lamina and the left lateral masses of C3, C4, C5 vertebrae) posterior stabilization was done with right side lateral mass screws fixation in C3 and C5 vertebrae. Core of the tumor consisted of cystic trabeculations containing unclotted blood and fleshy tissues.

After posterior surgery, the patient was made supine and through standard left anterior cervical approach, remaining part of the tumor was exposed. Anterior resection of the exophytic tumor followed by C4 corpectomy and C3-C4, C4-C5 discectomies done. Anterior spinal stabilization and fusion was done with an anterior cervical plate and mess cage packed with autologous iliac crest bone graft [Figures 4 and 5].
Patient was given soft cervical collar for 3 months postsurgery. The histopathology had confirmed the diagnosis of aneurysmal bone cyst. Patient is pain-free and there is no recurrence till now.

DISCUSSION

Aneurysmal bone cysts are benign, highly vascular, locally aggressive tumors, and recurrence rates after curettage were reported ≤50%.\(^1\) Spontaneous regression of the tumor is uncommon.

Aneurysmal bone cysts have a predilection for the lumbar spine in the series of Boriani et al.\(^1\) In contrast, in Papagelopoulos et al. and Vergel de Dios et al. series, cervical and thoracic spine was involved more than lumbar spine.\(^2\)

The combination of radiographs, CT scans, and MRI is diagnostic in many cases. Characteristic ballooning of the posterior elements with a thin rim may be seen on plain radiographs. CT imaging reveals multiloculated lytic lesions with multiple internal septations, pathological fracture or vertebral body collapse. CT scan is also useful for planning of possible instrumentation landmarks during surgery. On MRI, ABCs usually demonstrate a thin, well-defined rim of low signal intensity in the periphery, and they are seen as multiseptated heterogeneous lesions.

The histology of ABC is typically characterized by cavernous channels surrounded by a spindle cell stroma with osteoclast-like giant cells and osteoid production.

Differential diagnosis of ABCs includes giant cell tumor, simple bone cyst, osteoblastoma, chondroblastoma, chondromyxoid fibroma, fibrosarcoma, telangiectatic osteosarcoma, fibrous dysplasia, and plasmacytoma.\(^3\)

Many surgical, as well as nonsurgical management options, are given in the literature.

Nonsurgical management includes arterial embolization, radiation therapy, and intralesional steroid with calcitonin injection.

Although successful treatment with embolization has been given in aneurysmal bone cysts of pelvis and long bones but in spine, embolization has a very limited role especially with pathological fracture and neurological deficit where early surgical decompression is warranted.\(^4\) Preoperative embolization may be performed to minimize intraoperative blood loss.

Embolization may be considered as primary treatment in the patient with recurrent lesions after multiple surgery or inoperable lesion without any neurological deficit, pathological fracture and spinal instability, and deformity.

Role of percutaneous methylprednisolone and calcitonin intralesional injection has been described in the literature. Calcitonin inhibits osteoclastic activity and promotes trabecular bone formation while methylprednisolone has an antiangiogenesis action, but it has very limited role in the management.

There is controversy regarding the role of radiation therapy. Capanna et al.\(^7\) suggested that there was no benefit from the addition of radiation to partial resection and curettage. Patients in their series who received radiation therapy had poor outcomes: A delayed pathological fracture and kyphotic deformity developed in one; complications of cystitis, septicemia, and endocarditis in another; and a third experienced progression of disease that extended to adjacent levels resulting in quadriplegia.

Papagelopoulos et al. reported one case of postradiation osteosarcoma, which had occurred at the same site 7 years later, resulted in death.\(^4\)

Although curettage and bone grafting have been reported to be successful in the management of aneurysmal bone cysts in the long bones, the same does not apply to lesions of the spine. In a study by Ozaki et al.,\(^8\) 9 patients who underwent complete excision did not suffer a local recurrence, whereas two who underwent curettage alone experienced local recurrences of the tumor.

Total excision, en bloc if possible, provides the highest rate of cure with an excellent prognosis. Subtotal excision is associated
with a high incidence of recurrence, which is seen within 6-12 months. Hay et al.\(^1\) reported no recurrences after total excision and a 25% recurrence rate for partial excision.

Total excision must include the entire cyst as well as the abnormal surrounding tissue and the bony surfaces that are lined with the hypervascular membrane.\(^4,6,8\) If there is an extensive involvement of the vertebral bodies and posterior decompression is inadequate, then a separate procedure should be used whether in the same situation or at a subsequent stage.\(^4\) Widespread bone involvement can result in instability and extensive tumor resection can also cause postoperative iatrogenic instability. If instability and/or deformity exist or if the amount of bone removed would lead to instability, then simultaneous reconstruction and instrumentation are recommended.\(^3,4,6,8\) Posterior instrumentation using lateral mass screws or hooks and rods can be performed in the same instance.

If an anterior corpectomy is performed, then the anterior column needs to be reconstructed with an interbody graft and plating.\(^5,10\) This includes the anterior cervical plate for the cervical spine and the lateral interbody plate for the thoracolumbar region.\(^8\)

In cases in which postoperative instability is not obvious, we recommend a trial of external bracing with close radiographic follow-up. This is a reasonable alternative to instrumented fusion for children who have not reached skeletal maturity. If postoperative deformity develops, surgical stabilization is indicated.

**CONCLUSION**

Involvement of several adjacent cervical vertebrae by an aneurysmal bone cyst is very rare, and conventional treatment with curettage and bone grafting is most likely to carry a high rate of recurrence and spinal instability. We recommend complete excision of the tumor and instrumentation in a single stage to avoid instability.

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**Conflicts of interest**

There are no conflicts of interest.

**REFERENCES**