ScientificScholar®
Knowledge is power
Publisher of Scientific Journals

www.surgicalneurologyint.com

Surgical Neurology International

Editor-in-Chief: Nancy E. Epstein, MD, Clinical Professor of Neurological Surgery, School of Medicine, State U. of NY at Stony Brook.

SNI: Spine

Editor
Nancy E. Epstein, MD
Clinical Professor of Neurological Surgery, School of Medicine, State U. of NY at Stony Brook

SNI
Open Access

Case Report

A rare case of an expansile spinopelvic aneurysmal bone cyst managed with embolization, excision, and fusion

Rudra Mangesh Prabhu[1], Tushar N. Rathod[1], Akash Vasavda[1], Shivaprasad S. Kolur[1], Punit Tayade[2]

[1]Department of Orthopaedics, Seth G.S Medical College and K.E.M. Hospital, [2]Department of Physiotherapy, MGM School of Physiotherapy, Mumbai, Maharashtra, India.

E-mail: *Rudra Mangesh Prabhu - rmp31395@gmail.com; Tushar N. Rathod - tnrathod@gmail.com; Akash Vasavda - akashvasavda@gmail.com; Shivaprasad S. Kolur - shivaprasadkolur@gmail.com; Punit Tayade - punitstalion@gmail.com

***Corresponding author:**
Rudra Mangesh Prabhu,
Department of Orthopaedics,
Seth G.S Medical College and
K.E.M Hospital, Mumbai,
Maharashtra, India.

rmp31395@gmail.com

Received : 13 October 2021
Accepted : 05 November 2021
Published : 30 November 2021

DOI
10.25259/SNI_1045_2021

Quick Response Code:

ABSTRACT

Background: Aneurysmal bone cysts (ABC) are benign osteolytic lesions of the metaphyseal regions of long bones that typically contribute to rapid bony expansion. Here, we present an ABC involving the spinopelvic region in a 15-year-old male that required embolization, surgical excision, and fusion.

Case Description: A 15-year-old male, presented with gradually progressive painful lower back swelling of 4 months' duration. Once the diagnosis of an ABC was established based on a combination of X-ray, MR, and CT studies, he underwent selective arterial embolization, extended surgical excision (i.e. curettage), with a posterior fusion. Two years postoperatively, the patient remained neurologically intact without radiographic evidence of lesion recurrence.

Conclusion: Large expansile ABC involving the vertebral bodies should be managed with preoperative selective arterial embolization, surgical decompression/curettage, and spinopelvic fixation.

Keywords: Aneurysmal bone cyst, Embolization, Expansile, Spino-pelvic

INTRODUCTION

Aneurysmal bone cyst (ABC) primarily affects individuals between 10 and 20 years of age and is more common in females.[7,8] The treatment options include embolization, radiation therapy, simple curettage with or without bone grafting, complete excision, or a combination of these modalities.

CASE REPORT

Clinical presentation and radiographic findings

A 15-year-old male presented with gradually progressive painful low back swelling of 4 months' duration without any neurological deficit. Plain lumbosacral X-rays showed an enlarged lytic lesion involving predominantly the left side of the sacrum and lower lumbar vertebrae L4-S2 [Figure 1]. The MR demonstrated a large, multi-loculated, expansile mass with a soap-bubble-like appearance from L4-S2, which extended to the neural foramina, sacroiliac joints, and paravertebral muscles; findings were consistent with the diagnosis of an ABC [Figure 2]. The lumbosacral CT showed a lytic lesion involving the sacral alae, part of the S1and S2 vertebral bodies, and destruction of the left L5 pedicle [Figure 2].

Embolization of ABC lesion

An arterial angiogram confirmed the vascularity of the ABC mass. The patient underwent preoperative selective arterial embolization on the day of surgery, followed by an extended curettage [Figure 3]. This was followed by a posterior pedicle screw and rod lumbopelvic reconstruction (i.e. L4-S2) [Figure 4].

Pathology

The histopathological examination confirmed the diagnosis of an ABC lesion: osteoid foci, spindle cells, multinucleated giant cells, and reactive changes.

Postoperative follow up

Two years later, the patient remained asymptomatic without evidence of ABC lesion recurrence. The only focal asymptomatic finding on radiography was the loosening of the set screw on the left side inferiorly [Figure 5].

Figure 1: The preoperative antero-posterior and lateral radiograph of the spine. (a) The antero-posterior radiograph demonstrates an enlarged lytic lesion with predominant involvement of the left side of sacrum and lower lumbar vertebrae.

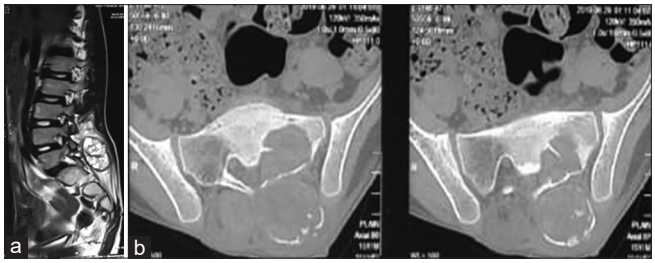

Figure 2: (a) T2-weighted MRI scan (sagittal cut) of the whole spine demonstrates 85 × 75 × 140 mm sized, large multi-loculated expansible mass with soap-bubble-like appearance occupying the region of L4, L5, S1, and S2. (b) CT scan (axial cut) shows a large lytic lesion involving both the sacral alae.

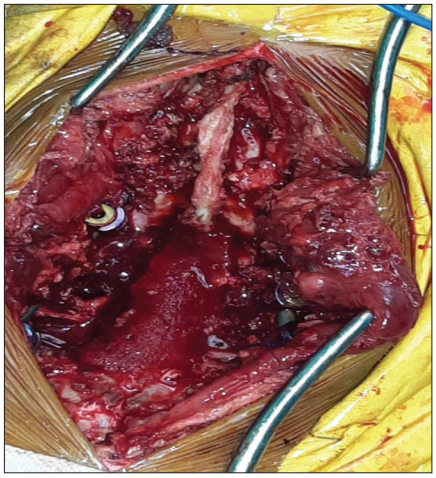

Figure 3: Intra-operative clinical image after extended curettage and decompression.

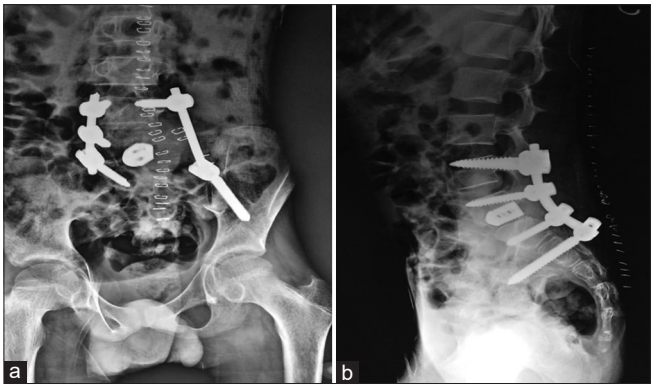

Figure 4: The post-operative anteroposterior and lateral radiograph of the spine. (a) Antero-posterior radiograph demonstrates bilateral pedicle screw fixation at L4, right-sided L5, and S1 pedicle screw fixation with a left trans-iliac pedicle screw and an anterolateral inter-body fusion at L5-S1 level. (b) The lateral radiograph demonstrates the screw and rod construct with the cage at the above-described levels.

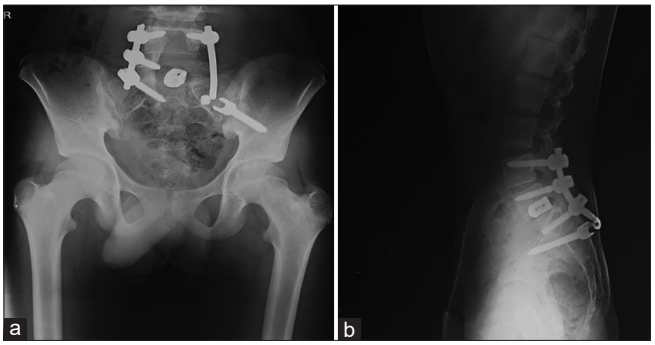

Figure 5: Follow up radiograph of the spine with the pelvis at 2 years post-surgery. (a) The antero-posterior radiograph shows the loosening of the set screw on the left side. (b) The lateral radiograph demonstrates loosening of the set screw inferiorly.

DISCUSSION

Clinical presentation and etiology of ABC lesions

ABCs have an inherent high local recurrence rate (i.e. 12 to 31.5%) even after "definitive" primary treatment.[9] The reappearance of pain or neurological deficit may indicate local recurrence of the lesion. Brembilla *et al*.[2] reported a cervical ABC lesion treated with aggressive surgical resection with instrumented fusion. Within nine postoperative months, the lesion recurred, following which the patient refused further treatment, and radiographs done at 5 years follow-up showed spontaneous cyst regression.

Frequency of ABC lesions

ABCs comprise 8–20% of all spine tumors with 3–12% occurring in the sacrum and pelvis, respectively.[6] Lumbar and thoracic vertebrae are common sites of involvement, with the sacrum being involved just 3% of the time.[6]

Pathogenesis of ABCs

Several theories have been proposed regarding the pathogenesis of ABCs. Preexisting pathology with secondary vascular factors may initiate the formation of a periosteal and/or intra-osseous arterio-venous malformation. Increasing hemodynamic forces generated by high-pressure vascular channels inside the cavity likely contribute to the rapid erosion of osseous trabeculae.

MR/CT DIAGNOSIS OF ABCS

MR findings

MRI best documents the extent of soft tissue involvement and neural compression. T2 studies usually show heterogeneous lesions containing multiple fluid-filled interfaces.[8] The administration of contrast further demonstrates internal septations within these cysts and helps differentiate them from other lesions (i.e., chordoma, metastatic disease).[8] This patient exhibited the classical MRI findings of multiple air-fluid levels, excavation/lytic changes of the L5 vertebral body, and an extensive soft tissue mass extending from L4-S2.

CT findings

With ABC lesions, CT scans of the spinal pedicles and vertebral bodies typically demonstrate multiple communicating cavities containing fluid-filled levels.

SELECTIVE PREOPERATIVE EMBOLIZATION

The main goal of selective arterial embolization in the management of ABCs is to decrease vascularity and reduce the level of intraoperative blood loss. Surgery is performed within 24 to 48 hours of embolization.[4,5] In our case, preoperative embolization was performed on the morning of surgery and there was minimal intraoperative blood loss.

ROLE OF RADIATION THERAPY

Radiation is not recommended as a primary treatment for the management of spinal ABC. Capanna *et al*. suggested that adjuvant radiotherapy alone had no significant advantages over surgical treatment alone; further, it might lead to an increased risk of malignant transformation, post-irradiation myelopathy, sarcoma, and growth disturbances in children.[3]

RECURRENCE OF ABCS

Incomplete resection of ABC at the time of the index surgery resulted in a 50 to 60% rate of recurrence within 6–12 months; recurrence after 2 years is unusual, and extremely rare after 4 years.[3,8] Hence, patients must be closely monitored postoperatively. Here, our patient was followed for 2 postoperative years without demonstrating recurrence. Radiological investigations performed at 2 years follow-up showed union at the L5-S1 level with no implant loosening, except for a set screw dislodgement on the left side. One of the plausible reasons for this was the predominant left-sided involvement by the tumor, leading to an excess loading on

Table 1: Literature review of sacral aneurysmal bone cysts.

Patient	Age in years	Sex	Management	Disease free follow-up (years)	Neurologic deficit
1	64	F	Resection	4	None
2	26	F	Curettage	2	None
3	39	M	Resection	2	Bowel and bladder dysfunction
4	16	F	Embolization with curettage	12	None
5	32	M	Curettage	5	None
6	60	F	Mid-Sacral Amputation	2	None
7	57	M	Embolization with mid-sacral amputation	2	Bowel and bladder dysfunction
8	10	F	Embolization with curettage	2	None
9	5	F	Curettage	2	None
10	17	F	Mid-Sacral Amputation	2	None

the left side. Initial post-operative non-compliance of the patient with bracing protocols could not be ruled out as a cause for the set screw loosening as the patient belonged to a rural agrarian background, which required a high level of ground-level activity.

REVIEW OF LITERATURE

Brastianos *et al*.[1] presented a review of 10 cases of ABCs of the sacrum [Table 1] managed with resection or curettage, of which 4 patients underwent preoperative embolization. Treatment results were excellent in their study.

CONCLUSION

Although ABCs are considered benign, they expand rapidly, have a high local recurrence rate, and can be mistaken as malignant tumors. Their management should include pre-operative selective arterial embolization, surgical excision/curettage, and stabilization.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

Financial support and sponsorship

Publication of this article was made possible by the James I. and Carolyn R. Ausman Educational Foundation.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Brastianos P, Gokaslan Z, McCarthy EF. Aneurysmal bone cysts of the sacrum: A report of ten cases and review of the literature. Iowa Orthop J 2009;29:74-8.
2. Brembilla C, Lanterna LA, Bosisio M, Gritti P, Risso A, Signorelli A, *et al*. Spontaneous regression after extensive recurrence of a pediatric cervical spine aneurysmal bone cyst. Case Rep Oncol Med 2014;2014:291674.
3. Capanna R, Albisinni U, Picci P, Calderoni P, Campanacci M, Springfield DS. aneurysmal bone cyst of the spine. J Bone Joint Surg Am 1985;67:527-31.
4. Chiras J, Cognard C, Rose M, Dessauge C, Martin N, Pierot L, *et al*. Percutaneous injection of an alcoholic embolizing emulsion as an alternative preoperative embolization for spine tumor. AJNR Am J Neuroradiol 1993;14:1113-7.
5. Gellad FE, Sadato N, Numaguchi Y, Levine AM. Vascular metastatic lesions of the spine: Preoperative embolization. Radiology 1990;176:683-6.
6. Han SR, Yee GT, Kim HS, Whang CJ. Aneurysmal bone cyst of a thoracic vertebra. J Korean Neurosurg Soc 2005;37:459-61.
7. Hong CK, Hyun DK, Park CO, Ha YS. A case of aneurysmal bone cyst on the thoracic spine. J Korean Neurosurg Soc 2000;29:675-9.
8. Liu JK, Brockmeyer, DL, Dailey AT, Schmidt MH. Surgical management of aneurysmal bone cyst of the spine. Neurosurg Focus 2003;15:E4.
9. Papagelopoulos PJ, Currier BL, Shaughnessy WJ, Sim FH, Ebersold MJ, Bond JR, *et al*. Aneurysmal bone cyst of the spine. Management and outcome. Spine (Phila Pa 1976) 1998;23:621-8.

How to cite this article: Prabhu RM, Rathod TN, Vasavda A, Kolur SS, Tayade P. A rare case of an expansile spinopelvic aneurysmal bone cyst managed with embolization, excision, and fusion. Surg Neurol Int 2021;12:580.