

Case Report

## Lumbar Spine Aneurysmal Bone Cyst: A Rare Entity

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### ABSTRACT

An Aneurysmal Bone Cyst (ABC) is a benign, locally aggressive, vascular, and expansile bony tumor of idiopathic etiology containing multiple thin-walled blood-filled channels, mostly diagnosed in pediatric and adolescent age groups. These lesions can cause local pain, pathological fractures, spinal deformity, and neurological deficits. The treatment of choice for ABC is highly debatable according to the literature. The treatment choices are simple curettage and grafting, complete surgical resection with or without prior selective arterial embolization, radiotherapy, or a combination of these procedures according to the case. Each modality is having different outcomes, technical requirements, and complications. We are reporting a case of Aneurysmal Bone Cyst of the lumbar spine in a young patient treated by surgery.

**Keywords:** Aneurysmal Bone Cyst (ABC), Selective Arterial Embolization, Lumbar Spine, Surgical Resection.

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### INTRODUCTION

An Aneurysmal Bone Cyst (ABC) is benign but locally aggressive, idiopathic, hemorrhagic, and multicystic bone tumor containing multiple thin-walled blood-filled channels of variable size, not lined by endothelium, separated by connective tissue containing trabeculated bone tissue and osteoclast giant cells. These are mostly diagnosed

in pediatric and adolescent age groups, representing 1.4 to 2.3% of primary bone lesions,<sup>1</sup> 80% found in juvenile patients of age less than 20 years.<sup>2</sup> Although often primary, up to 1/3<sup>rd</sup> of ABCs are resulting from a pre-existing underlying bone pathology e.g. fibrous dysplasia, giant cell tumor of bone,<sup>3</sup> osteosarcomas, chondromyxoid fibroma,<sup>5</sup> non-ossifying fibroma,<sup>6</sup> chondroblastoma.<sup>7</sup> Most commonly found in the locations as listed in Table 1.<sup>3,4,8</sup>

The lesion in the spine usually presents with local pain, pathological fractures, spinal deformity, and neurological deficits. If we study the literature, different authors have different opinions regarding the treatment of choice for ABC. The treatment choices include simple curettage and grafting, complete surgical resection with or without prior selective arterial

**Table 1:** Skeletal Distribution of Aneurysmal Bone Cyst.

Parts of Body	Location	Percentage %	
Long Bones	Upper Limbs	10 – 20	
	Lower Limbs	Femur	13
		Tibia/Fibula	24
Spine	Cervical Spine	2	
	Thoracic Spine	8 – 10	
	Lumbar Spine	15 – 20	
Head & Face	Mandible	10-20	
	Basisphenoid		
	Paranasal Sinuses		

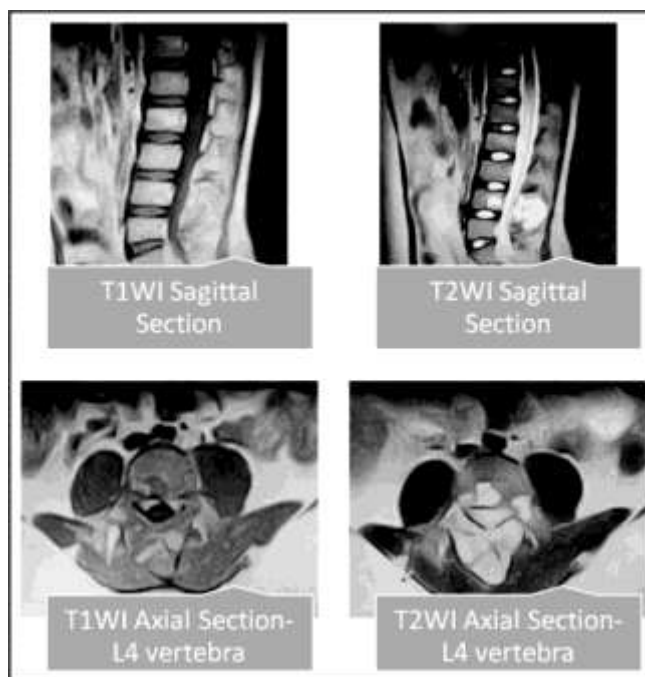
embolization (SAE), radiotherapy, or using a combination of these procedures depending upon the case, with each procedure having different outcomes, technical requirements, and complications. The treatment of choice in case of a highly aggressive tumor in the spine is complete surgical resection of the tumor with or without spinal stabilization by fixation depending upon the case. There is an increased risk of bleeding as these are highly vascular lesions. We are reporting a case of Aneurysmal Bone Cyst of the lumbar spine in a juvenile patient treated by surgery, as the literature shows it is a rare case and there is very limited research available on Aneurysmal bone cysts nationally.

**CASE REPORT**

A 15 years old Male patient presented in Outpatient Department of Neurosurgery at Sharif Medical City Hospital on 18-12-2020, with complaints of low back pain radiating to both legs, progressive bilateral lower limb weakness, and difficulty walking for 2 years. The patient had a preceding history of falls from stairs after their foot slipped, 2 years back. The patient also had a complaint of low back pain during micturition. No history of urinary or fecal incontinence. On basis of 11 point-Numeric Pain Rating Scale (0-no pain to 10-worst pain), pt. recorded pain at 8 points (severe). On Neurological Examination, the

patient had weakness of bilateral lower limbs with Power 4/5 in both lower limbs mainly involving L3, L4, and L5 myotomes (weakened quadriceps, knee extensors, ankle dorsiflexors, and long toe extensors). Straight Leg Raising test (SLR) of 10 degrees in right and left lower limbs. Decreased pain and touch sensations at L4 and L5 dermatomes on the left side with intact sensations in all other dermatomes. Normal Anal tone and perianal sensations. Plantar reflex bilateral down going.

MRI Lumbosacral spine showed expansion of posterior elements (involving spinous process, both pedicles, and lamina) of L4 vertebra and occupied by well-defined multiloculated altered signal intensity cystic lesion measuring 4x4.3x4.7cm (CC, AP & Transverse dimension). The lesion was iso-to-hypo intense on T1WI and hyperintense on T2WI. The fluid-Fluid level was seen in some cysts. After Intravenous contrast,



**Figure 1:** Preoperative MRI Lumbosacral Spine showing heterogeneous mass lesion involving posterior elements of L4 vertebra expanding to the vertebral body, iso-to-hypointense on T1WI and Hyperintense on T2WI, with the fluid-fluid interface.

patchy enhancement was seen. There was also thecal sac and nerve roots compression (Figure 1).

On the basis of the MRI Lumbosacral spine, the differential diagnosis was made of primary and secondary bone lesions like simple bone cysts, giant cell tumor, aneurysmal bone cyst, chondroblastoma, osteosarcoma etc.

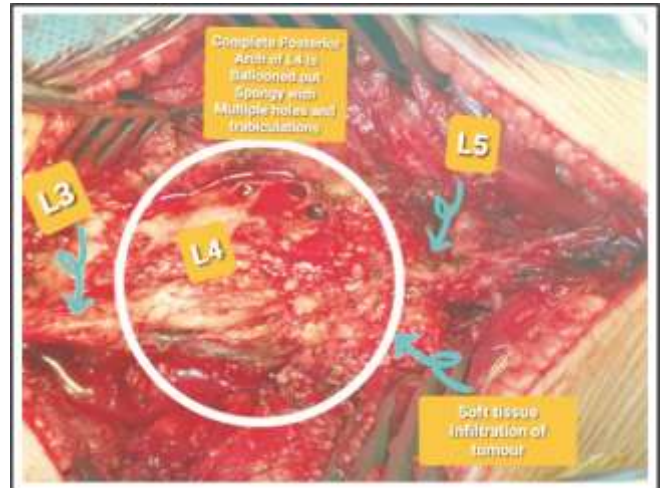
The surgery was performed for excision of this lesion using the posterior approach on 21-12-2020. Perioperative Fluoroscopy was used to reconfirm and identify the exact location of the tumor as shown in (Figure 2).



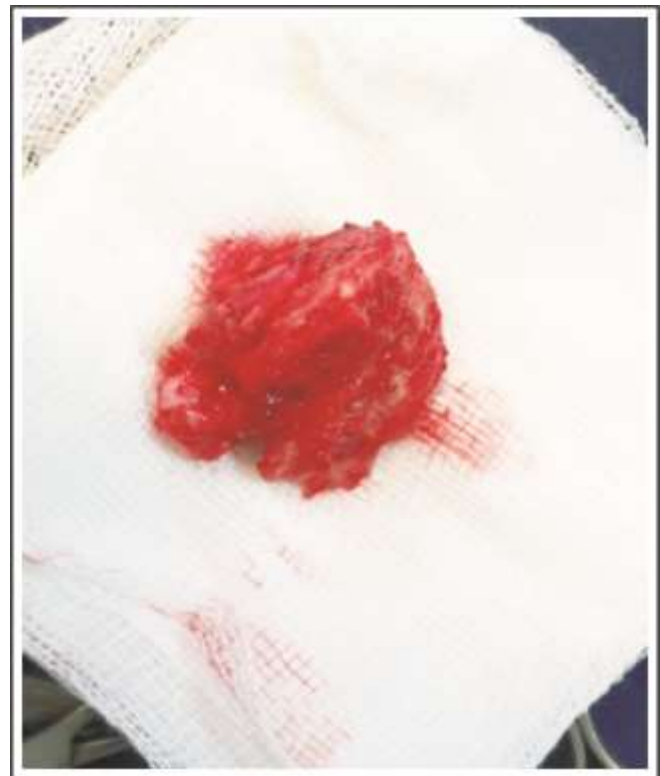
**Figure 2:** Perioperative Fluoroscopy of Lumbar spine showing Space Occupying Lesion (SOL) involving complete L4 vertebral Posterior arch represented with a marker in place and partially L3 and L5 vertebral posterior arches.

Operative Findings included: Vertebral deformity involving posterior elements of L3 (lower border), L4, and partially L5 vertebrae. Multiple holes found in the cortex of the L4 vertebra posteriorly on either side. Ballooned out bony structure with multiple septa of spongy bone expanding the posterior element anteriorly and posteriorly causing spinal canal compression and bilateral neural foramina stenosis. The tumor

was pinkish in color and highly vascular (Figures 3, 4).

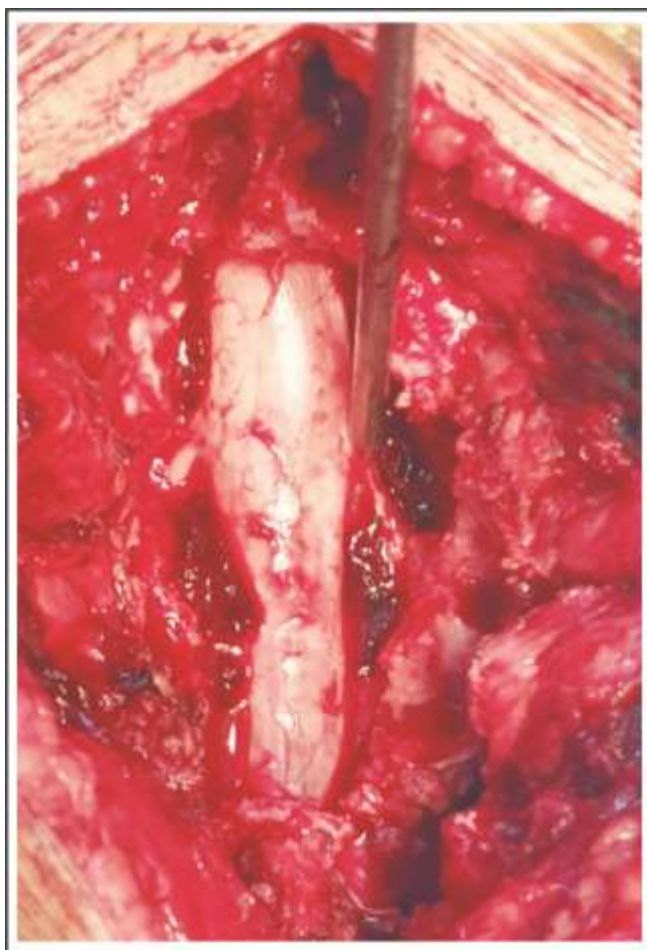


**Figure 3:** Perioperative post-exploration view showing ballooned out the spongy bone with multiple trabeculations involving complete posterior elements of L4, lower border of L3 and Partially L5 vertebrae.



**Figure 4:** Tumor Biopsy specimen.

Complete surgical resection of the tumor from the lower border of L3 to partial L5 level with complete decompression of the thecal sac, bilateral foramina, and nerve roots, complete removal of bony spicules, and securing of hemostasis was done (Figure 5). Adequate blood was arranged preoperatively for anticipated hemorrhage. The tumor was excised in en bloc fashion (instead of piecemeal fashion) to minimize bleeding and to effectively secure hemostasis peroperatively.



**Figure 5:** Peroperative view showing complete excision of Space Occupying Lesion with complete decompression of thecal sac and bilateral nerve roots.

Histopathology report showed multiple dilated blood filled cystic spaces lined by

multinucleated giant cells with fibroblastic proliferation and osteoid formation, without the presence of atypia, confirmed Aneurysmal Bone Cyst.

The patient was followed up after 2 weeks and 6 weeks. After 2 weeks' follow-up, the intensity of pain decreased, according to numeric pain rating scale, from (preoperative) 8 points (severe) to (post-operative) 2 points (mild). Post-operative X-ray Lumbosacral spine (Anteroposterior and Lateral view) was done at 2 weeks follow up, showing excision of tumor from posterior elements at L3, L4, and L5 level with no disco-vertebral deformity (Figure 6).



**Figure 6:** Post-operative X-ray Lumbosacral spine having Anteroposterior (A) and lateral (B) views, showing Excision of Tumor from posterior elements of L3, L4, and L5 vertebrae with no disco-vertebral instability.

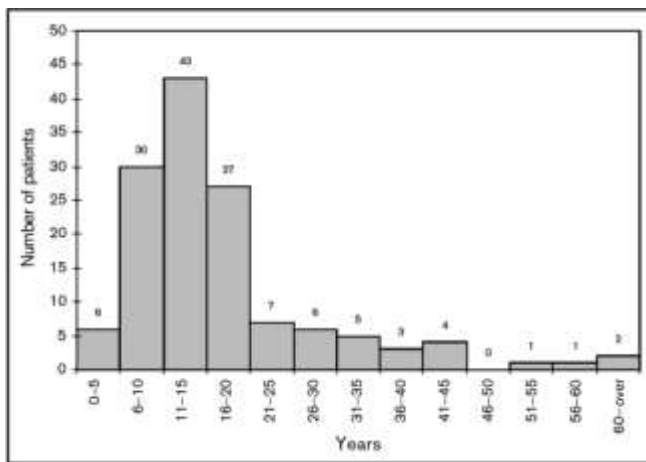
At 6 weeks' follow-up, pt. was having no pain at back or in legs and marked improvement in other symptoms as well with no weakness or numbness of any part of legs and was able to walk easily with no support.

The patient was referred to Oncology Department for consideration of post-surgical radiation.

## DISCUSSION

Aneurysmal bone cysts (ABCs) were first described by Dr. Jaffe and Dr. Lichtenstein in

1942. ABCs are not lined by endothelium and are neither true aneurysmal nor cystic so their name is a misnomer. Aneurysmal Bone Cysts are benign, highly vascular, expansile, locally reactive, and proliferative bony tumors of unknown etiology. Aneurysmal bone cysts contain multiple thin-walled blood-filled channels of variable size, separated by a layer of connective tissue containing trabeculated bone tissue and osteoclast giant cells. It usually affects the pediatric population, 80% found in young patients of age less than 20 years, as presented in (Figure 7). Females are affected predominantly (F:M ratio is 1.3:1).<sup>2,21</sup>



**Figure 7:** Demographic data for 135 patients with Aneurysmal bone cyst.

Although 50 – 60% of ABCs occur in the metaphysis of long bones, about 20% to 30% of cases occur in the spine especially in posterior elements of spine<sup>3</sup> with up to 40% of these are extended into pedicles, vertebral bodies and spinal canal causing pathological fractures and neurological deficits.<sup>4</sup> ABCs most commonly involve the lumbar area in the spine.<sup>8</sup>

Patients with Lumbar ABCs usually present with low back pain, stiffness in lower limbs, swelling, weakness of limbs, paresthesia, and bone fractures.<sup>9,10</sup> Most of these lesions appear small, solitary and involve a single segment of the

spine.<sup>11</sup> The diagnosis and surgical planning of ABCs require preoperative radiological assessment using CT scan and MRI.<sup>12</sup> The distinguishing traits of ABCs are better observed on MRI,<sup>13</sup> showing the presence of multiloculated cystic channels with fluid-fluid interfaces, on T1- and T2-weighted images having varying signal intensity most likely representing areas of the blood of different ages.<sup>14</sup> On MRI T1WI with contrast, septations may enhance.<sup>14</sup> However, the presence of fluid-fluid interface is also seen in other bone lesions as well like giant cell tumors (GCT), simple bone cysts, chondroblastoma, and telangiectatic osteosarcomas. So these lesions must be included in the differential diagnosis of ABCs.<sup>15</sup> The definitive diagnosis is confirmed by biopsy of the tumor/lesion.

ABCs are mostly treated surgically. Preoperative selective arterial embolization (SAE) can be helpful in the identification of feeding vessels to minimize intraoperative blood loss which is required in some cases.<sup>13</sup> The possible postoperative complications after surgery may include neurological deterioration or deficit, massive hemorrhage, infection, and recurrence of the tumor.<sup>16</sup> These can be prevented by wide operative field exposure and preoperative SAE. Complete surgical excision of the lesion along with its wall reduces the risk of recurrence. Radiotherapy is not considered to be the first-line treatment option for spinal ABC, in view of its potential complications and risks of post-radiation growth restraint, myelopathy, spinal deformity, and malignant transformation like sarcoma, especially in children.<sup>12,13,17-20</sup> Radiotherapy is usually used after surgery or in patients who are unfit for surgery.

In this case, we performed complete exposure and complete en bloc surgical resection of the tumor using the posterior approach with complete decompression of thecal sac, bilateral foramina and nerve roots, complete removal of bony spicules, and securing of hemostasis from the lower border of L3 to partial L5 level.

## CONCLUSION

ABC of the lumbar spine is a benign, highly vascular, and locally aggressive primary bone lesion with a high rate of recurrence. Treatment choices for lumbar ABC include Curettage and bone grafting, complete surgical excision with or without pre-operative Selective Arterial Embolization, and radiotherapy which can be used alone or in combination. The recurrence of ABC can be avoided by wide exposure and complete surgical excision of ABC along with its entire wall, with special consideration to the adolescent population to minimize the risk of post-surgical spinal deformity and neurological deterioration.

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### Additional Information

**Disclosures:** Authors report no conflict of interest.

**Ethical Review Board Approval:** The study was conformed to the ethical review board requirements.

**Human Subjects:** Consent was obtained by the patient in this study.

**Conflicts of Interest:**

In compliance with the ICMJE uniform disclosure form, all authors declare the following:

**Financial Relationships:** All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work.

**Other Relationships:** All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

### AUTHORS CONTRIBUTIONS

Sr.#	Author's Full Name	Intellectual Contribution to Paper in Terms of:
1.	Babar Butt	Operating Surgeon and Primary Author.
2.	Syed Ali Zunair	Assistant Surgeon, Literature review, data interpretation and manuscript writing.
3.	Alia Latif	Data collection, Pre-operative, post-operative and management of patient.
4.	Nasir Raza Awan	Review of Manuscript.