

Case Reports & Case Series

Aneurysmal bone cyst of the spine: Report of four cases and review of the literature

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A B S T R A C T

Purpose: We report four cases of spinal Aneurysmal bone cyst (ABC) that were managed surgically at King Fahad Specialist Hospital in Dammam, Saudi Arabia. This report aims to describe the clinical features and the need for surgical excision preceded by embolization in spinal ABC.

Method: A retrospective review of the OR list and tumor board records between 2013 and 2016 yielded a total of 4 cases of spinal ABC who were surgically treated at KFSSH-D. Their clinical presentations, radiological and pathological features are reviewed along with the surgical intervention utilized and its outcome.

Results: Total of 4 cases were identified, all of them were female (100%). The age ranged from 13 to 25 years old (mean age = 17.75). Two cases (50%) were located in the thoracic spine, one (25%) cervical and one (25%) in the lumbar spine. Back pain, unsteady gait, and progressive limb weakness were the most common presenting symptoms. All the cases underwent embolization followed by surgical excision and fixation with a variable degree of improvement postoperatively.

Conclusion: Spinal ABC is a benign tumor that can present with a wide variety of non-specific symptoms. Pre-operative embolization can be helpful to reduce bleeding during surgery. Surgical decompression can lead to significant improvement even in case of severe neurological deficit. Radiation therapy can be used as an adjunct treatment in selected cases.

1. Introduction

Aneurysmal bone cyst (ABC) is an expansile, non-neoplastic lesion that contains several blood-filled endothelialized cavities. This lesion was first time described by Jaffe and Lichtenstein 1942. They described a lesion that appeared to be a blood-filled cavity that had a blowout radiographic appearance analogous to a saccular aneurysm. [1] The etiology of an aneurysmal bone cyst is uncertain. It is hypothesized that the aneurysmal bone cyst is reactive, non-neoplastic process that has the ability to destroy and expand bone.

It may occur in bone as a solitary lesion or can be found in association with other bone tumors such as giant cell tumor, chondroblastoma, chondromyxoid fibrous dysplasia, or in association with a malignant process [2]. ABC is a rare bone tumor representing approximately 1% of all bone solid tumor with an annual incidence of 1.4 in 100,000 people [3]. More than two-thirds of the ABC occurs below the age of 20 [4]. It frequently involves the metaphysis of the long bones mainly around the knee. Approximately one-third of these tumors arise from the spine. They represent approximately 15% of all

primary spine tumors [5]. Managing spinal aneurysmal bone cyst can be challenging. Treatment can be achieved by surgical resection, radiation therapy, arterial embolization or combination of these modalities [6].

2. Method

A retrospective analysis of the operation list and tumor board records for the years 2013, 2014, 2015 and 2016 resulted in total of four cases of ABC involving the vertebral column that were offered surgical management. Their clinical presentations, radiological and pathological features are reviewed along with the surgical intervention utilized and its outcome.

3. Results

Total of four cases was identified; all cases were female (100%). The age ranged from 13 to 25 years old (mean = 17.75). Two cases (50%) were located in the thoracic spine, one (25%) in the cervical and one

Abbreviations: ABC, aneurysmal bone cyst; OR, operative room

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Fig. 1. Case no. 1: C7 shows an expansile mass which involves the transverse process, pedicle and lamina. It crosses midline into the anterior part of the vertebral body of C7. There is an extra osseous extension with involvement of the epidural space extending from C5-T1 causing compression of the cord and deviation to the left. C7 corpectomy, tumor debulking + C6-T1 expandable cage and graft + plate fixation + neck soft tissue dissection + posterior decompression and C6 lateral mass screw, T1 pedicle screw.

(25%) in the lumbar spine. Presenting symptoms were: Pain (75%) unsteady gait (75%), lower limb weakness (50%), radiculopathy (25%) and local back swelling (25%). Spastic paraparesis was present in 2 patients (50%) and decreased sphincter tone in one patient (25%).

Radiologically, all patients had posterior elements involvement and three patients had a variable degree of cord compression. The cervical ABC was found to be secondary to giant cell tumor and it was involving the right transverse process, pedicle, lamina and anterior part of the vertebral body of C7 with an extra osseous extension and vertebral artery encasement (Fig. 1). The two thoracic ABC cases (T8 and T10) had a posterior element with part of the vertebral body involved with evidence of cord compression (Figs. 2 and 3). The case with lumbar ABC had complete involvement of the L2 vertebral body and its posterior elements including the spinous process (Fig. 4). All patients underwent preoperative selective arterial embolization before definitive surgery. One case with thoracic ABC at the level of T8 developed acute lower limb paraplegia few hours after the embolization which prompted an emergent decompression and fixation. All the patients had surgery in form of curettage and bone grafting. Surgical decompression and tumor resection through posterior approach with instrumentation was used for the four cases. The anterior approach was also used in the cervical spine. Complete resection of the lesion was achieved in the 3 cases

(75%). Post-operative radiotherapy was not used in any case. The follow-up period is relatively short (mean 2.5 years) and so far no evidence of radiological recurrence was observed (Table 1).

4. Discussion

Aneurysmal Bone Cyst is non-neoplastic expansile bone lesion that consists of blood-filled spaces separated by connective tissue septa containing bone trabeculation and osteoclastic giant cells [19]. The prevalence of ABCs is estimated at 1.4 per 100,000, representing almost 1% of all bone tumors. The lesions slightly affect females more than males and mainly occur in the first two decades of life [3,5]. In our report, all of the patients are females with the mean age of 17.75 years similar to the literature. In his report of 41 cases, Boriani et al. [5] showed that the lumbar spine was frequently involved (18 cases, 45%). In contrast, other reports showed that the cervical and thoracic spine are more frequently involved [6,8]. In our report, the thoracic spine was involved in 50% of the cases. Back Pain was the most common presenting symptom in our report; however, neurological manifestations are not uncommon. These manifestations include limb weakness, ataxic gait and bowel or bladder dysfunctions.

On Imaging, plain radiograph usually show a ballooning of the

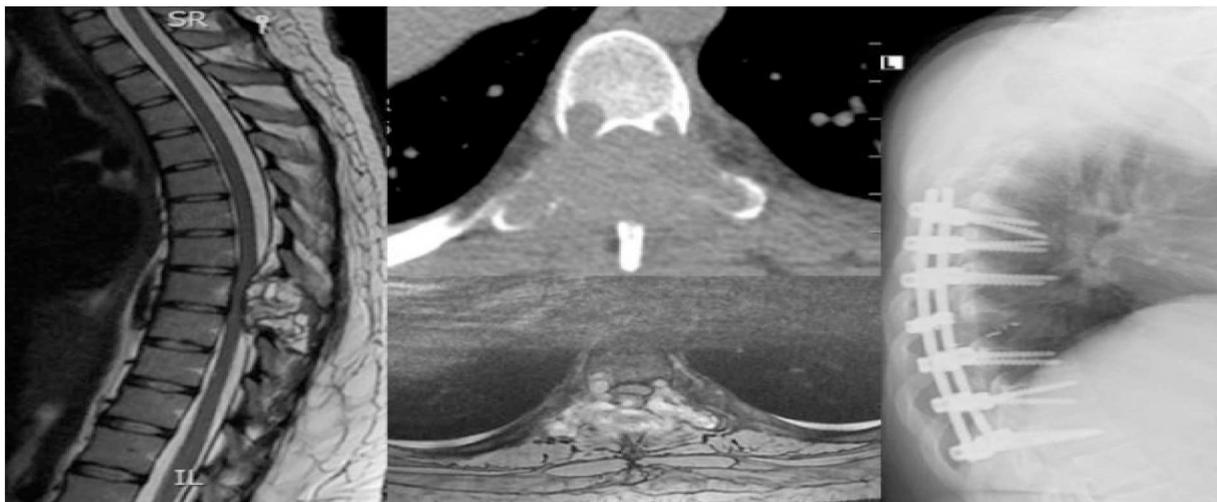


Fig. 2. Case no. 2: T8 mainly posterior arch lytic lesion eroding lamina, pedicle, spinous process and minor part of the T8 with cord compression. Posterior approach total ABC excision with T5–T11 pedicle screws fixation.

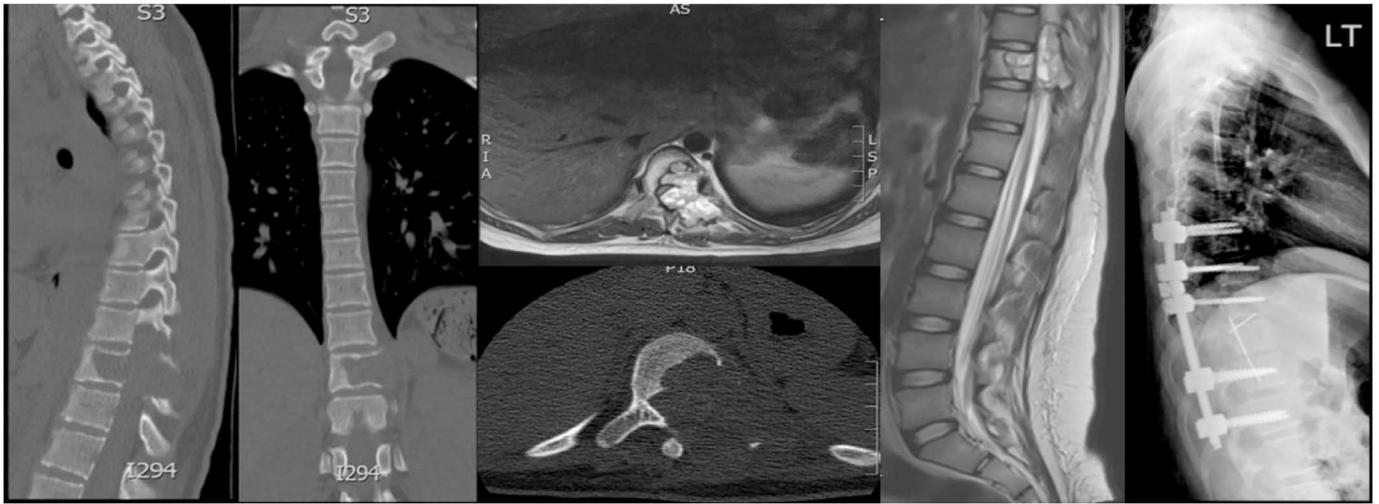


Fig. 3. Case no. 3: T10 lobulated lytic lesion with faint post IV contrast enhancement and few fluid-fluid levels. ABC involving mainly posterior middle aspect of T10 and left sided lamina and pedicle including the superior and inferior articular process and part of the spinous process with evidence of cord compression. Left T10 costotransversectomy + T9–10 laminectomy, fixation T7–12.

vertebrae with thin bony rim. Computed Tomography can show a pathological fracture of the thin bone cortices. MRI is the imaging modality of choice for the diagnosis of the ABC. MRI of the ABC has a characteristic of Multiseptated, well-defined cystic lesions surrounded by a rim of low signal intensity that contains heterogeneous signal intensity on both T1 and T2 weighted images. ABC might have a fluid-fluid level but this finding is not specific as it can be seen in several other pathologies such as osteosarcoma and giant cell tumor [9]. Both CT and MRI are helpful in preoperative planning.

Histopathological examination of ABC is described as cavernous spaces and blood-filled bony trabeculation that can be lined with giant cells, spindle cells and reactive bone formation [10,18]. Some theories suggest that the tumor is secondary to bleeding into a tumor or repair of subperiosteal hemorrhage [9].

Treatment of the spine ABC is controversial, with surgical and non-surgical options available [19]. Non-surgical options include arterial embolization and calcitonin or steroid intralésional injection. Radiotherapy can be utilized if the lesion deemed unresectable or in case of incomplete resection as an adjuvant therapy, however, it carries risk for complications such as osteonecrosis and myelopathy [14,15]. Options for surgical treatment are either en bloc resection or intralésional curettage with bone grafting [5–11]. Zileli et al. in his report of 13 patients reported the application of Polymethyl Methacrylate (PMMA) for

vertebroplasty/kyphoplasty in 4 patients to further reinforce spine stability post-resection [18].

Complete surgical resection is the optimal approach for local tumor control and recurrence prevention. Instrumentation can also be considered as surgical intervention might lead to spinal instability [12,13]. As the technique of complete resection of the ABC is reported to be challenging [5] all our patients had intralésional curettage and due to the high possibility of spinal instability in large lesions resection, instrumentation was also performed.

Intraoperative bleeding is common in ABC. Preoperative selective embolization should be considered to reduce the risk of intraoperative bleeding [5,16]. All our patients underwent embolization preoperatively, therefore, it was difficult to compare the significance of blood loss with or without preoperative embolization. Our patient with T8 ABC developed a complication in form of acute paraplegia following the embolization that improved partially post-operatively.

Recurrence is seen in 10–44% of the cases. Complete excision help reduce the risk of recurrence [8]. Zenonos reported two ABC that recurred after a failure of complete en bloc resection which occurred 9 months to 8 years after surgical intervention [13]. Follow up after surgery for 2 years is recommended as 90% of recurrences occur within this period, the follow-up period can be extended in case of intralésional curettage was conducted and in case of incomplete resection

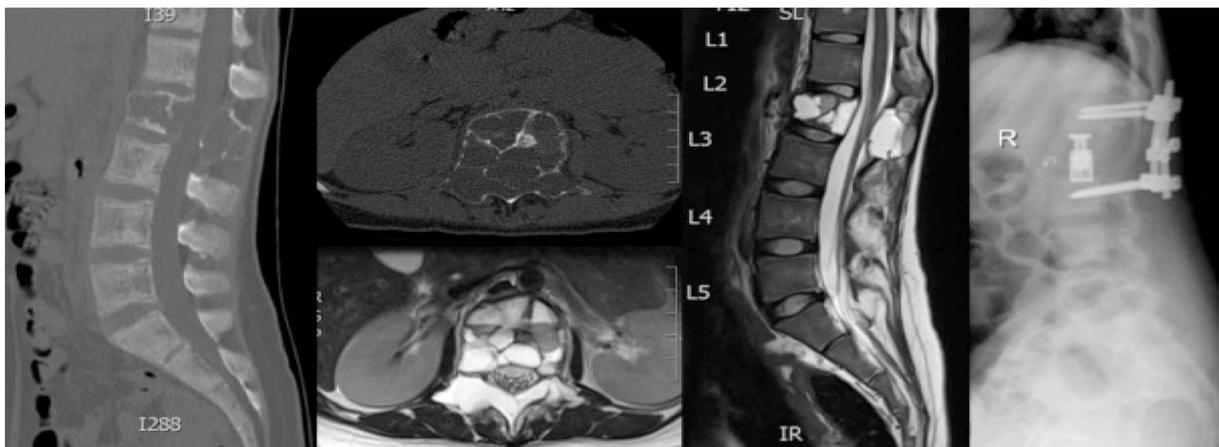


Fig. 4. Case no. 4: L2 is multiseptated expansile lytic lesion with remodeling of cortical margin involving the whole vertebral body of L2 extending to the pedicle to the spinous process with multiple bony septation associated with expansion and focal kyphosis. ABC excision and L1–L3 posterior fixation and cage insertion.

Table 1
Summary of the cases.

Case	Level	Age/gender	Main findings	Radiology	Posterior elements involvement	Outcome	Recurrence
1	C7	16/F	Radiculopathy/mild weakness	Expansile mass. Giant cell tumor	Yes	No improvement from baseline	No
2	T8	25/F	Spastic weakness/ataxia/incontinence	Mainly posterior elements/cord compression	Yes	Acute paraplegia post embolization/improved mildly post-operatively	No
3	T10	13/F	Spastic weakness/ataxia	Fluid-fluid levels/cord compression	Yes	Motor power partially improved	No
4	L2	17/F	LBP/radiculopathy/back bulge	Complete replacement of L2/kyphosis/canal stenosis	Yes	Partially improved/no deficit developed	No

[5,13,17]. We have an average follow up period of 2.5 years and so far, no recurrence detected.

5. Conclusion

ABC is a benign tumor that can present with a wide variety of non-specific symptoms. Neurological deficits are not uncommon. Pre-operative embolization can be helpful to reduce bleeding during surgery. Early diagnosis and careful surgical planning is essential. Surgical decompression can lead to significant improvement even in case of severe neurological deficit.

Disclosure

The authors declare that there are no conflicts of interest regarding the publication of this paper.

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References

- [1] J.L. JaVe, L. Lichtenstein, Solitary unicameral bone cyst with emphasis on the roentgen picture, *Arch. Surg.* 44 (1942) 1004–1025.
- [2] M.R. Brinker, T.S. Whitecloud, M.R. Hand, et al., Vertebral aneurysmal bone cyst. A case report and review, *Orthop. Rev.* 20 (1991) 42–46.
- [3] A. Leithner, R. Windhager, S. Lang, O. Haas, F. Kainberger, R. Kotz, Aneurysmal bone cyst. A population based epidemiologic study and literature review, *Clin. Orthop. Relat. Res.* 363 (1999) 176–179.
- [4] M.A. Blake, *Imaging in Oncology*, Springer Verlag, 0387755861, 2008.
- [5] S. Boriani, F. De Iure, L. Campanacci, et al., Aneurysmal bone cyst of the mobile spine: report on 41 cases, *Spine* 26 (2001) 27–35.
- [6] P.J. Papagelopoulos, B.L. Currier, W.J. Shaughnessy, et al., Aneurysmal bone cyst of the spine. Management and outcome, *Spine* 23 (1998) 621–628, <https://doi.org/10.1097/00007632-199803010-00018>.
- [7] F. Schajowicz, *Histological Typing of Bone Tumors (International Histological Classification of Tumors)*, Springer, New York, NY, USA, 1993.
- [8] A.M. Vergel de Dios, J.R. Bond, T.C. Shives, R.A. McLeod, K.K. Unni, Aneurysmal bone cyst. A clinicopathologic study of 238 cases, *Cancer* 69 (1992) 2921–2931.
- [9] P.L. Munk, C.A. Helms, R.G. Holt, J. Johnston, L. Steinbach, C. Neumann, MR imaging of aneurysmal bone cysts, *AJR Am. J. Roentgenol.* 153 (1) (1989) 99–101.
- [10] G. Al-Shamy, K. Relyea, A. Adesina, W.E. Whitehead, D.J. Curry, T.G. Luerssen, A. Jea, Solid variant of aneurysmal bone cyst of the thoracic spine: a case report, *J. Med. Case Rep.* 5 (261) (2011) 1–6.
- [11] P. Tsagozis, O. Brosjö, Current strategies for the treatment of aneurysmal bone cysts, *Orthop. Rev. (Pavia)* 7 (4) (2015) 6182.
- [12] R. Saccomanni, Aneurysmal bone cyst of spine: a review of literature, *Arch. Orthop. Trauma Surg.* 128 (2008) 1145–1147.
- [13] G. Zenonos, O. Jamil, L.S. Governale, et al., Surgical treatment for primary spinal aneurysmal bone cysts: experience from Children's Hospital Boston, *J. Neurosurg. Pediatr.* 9 (2012) 305–315.
- [14] J.C. Tsai, M.K. Dalinka, M.D. Fallon, M.B. Zlatkin, H.Y. Kressel, Fluid–fluid level: a nonspecific finding in tumors of bone and soft tissue, *Radiology* 175 (3) (1990) 779–782.
- [15] J.N. Weinstein, R.J. McLain, Primary tumors of the spine, *Spine* 12 (1987) 843–851.
- [16] G. Rossi, E. Rimondi, T. Bartalena, A. Gerardi, M. Alberghini, E.L. Staals, C. Errani, G. Bianchi, A. Toscana, M. Mercuri, D. Vanel, Selective arterial embolization of 36 aneurysmal bone cysts of the skeleton with N-2-butyl cyanoacrylate, *Skelet. Radiol.* 39 (2010) 161–167.
- [17] D.J. Ruiters, T.G. Van Rijssel, E.A. Van Der Velde, Aneurysmal bone cysts: a clinicopathological study of 105 cases, *Cancer* 39 (1977) 2231–2239.
- [18] M. Zileli, H. Isik, F. Ogut, M. Is, S. Cagli, C. Calli, Aneurysmal bone cysts of the spine, *Euro Spine J.* 22 (2013) 593–601.
- [19] S. Ozdemir, C. Yaldiz, F. Ozden, O.K. Kacira, T. Kacira, Aneurysmal bone cysts of the spine: two case reports, *Kor. J. Spine* 11 (4) (2014) 249–251, <https://doi.org/10.14245/kjs.2014.11.4.249>.