

Aneurysmal Bone Cysts of the Spine

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Aneurysmal bone cysts (ABCs) were originally described by Jaffe [1] and Lichtenstein [2] in 1942 and further characterized by others [3]. The term “aneurysmal bone cyst” is a descriptive term describing the pathologic appearance of the lesion within bone. It has been described as bone with “blown out distension with blood filled cavities” or as “having the appearance of a blood soaked sponge” [4]. It has been defined by the World Health Organization as “an expanding lesion with blood filled cavities separated by septa of trabecular bone or fibrous tissue containing osteoclast giant cells.”

ABCs compromise 1% to 6% of solid bone tumors [5,6]. The lesion is usually found in the femur, tibia, humerus, and spine [7]. ABCs have also been rarely reported in the small bones of the hands and feet [3]. ABCs represent approximately 15% of all primary spine tumors, with 8% to 30% of all ABCs arising in the spine [4,7]. In the mobile spine almost all of the lesions involve the posterior elements, although the lesions characteristically often also involve the vertebral body [4]. In Boriani and colleagues [4] and Collordata and colleagues [7] series the lumbar spine was most affected (40%–45%), with lesions in the cervical (30%) and thoracic spine (25%–30%) occurring less commonly.

Overall ABCs are more common than osteosarcomas and giant cell tumors in the spine. Leithner and colleagues [5] estimated the annual incidence of the disease as 0.14/10,000 per population. The true incidence of the disease is difficult to determine, however, because some lesions

remain benign and regress spontaneously with or without biopsy [8,9], whereas others have been diagnosed incidentally. Because of the rarity of the condition, the literature regarding ABCs consists of mainly case reports and small retrospective clinical series.

Classification and pathology

ABCs are benign lesions and can be classified according to Enneking as type 1 (latent), type 2 (active), and type 3 (aggressive) lesions. Furthermore, ABCs can be classified as primary or secondary where primary lesions are defined as those not associated with other lesions. In one third of cases, however, ABCs are secondary; they are most commonly associated with giant cell tumors, hemangiomas, osteblastomas, chondroblastomas, and telangiectatic osteosarcomas [10]. The secondary association makes the diagnosis and treatment in some cases difficult [11]. A solid variant has also been described that accounts for 3.4% to 7.5% of all ABCs. It differs in that cavernous channels and spaces are absent, yet the histology remains consistent and characteristic for an ABC [4,6,12–14]. Like primary and secondary ABCs, the solid variant contains histioblasts, giant multinucleated cells, thin capillary vessels, hemosiderin granules, and immature trabecular bone [4,15].

The pathophysiology of ABCs remains unclear and is most likely due to a non-homogeneous process. Lichtenstein proposed that an ABC is a reactive lesion produced through a vascular disturbance in bone that leads to increased venous and osseous pressure causing a local distension of bone. Biesecker and colleagues performed manometric tests on six ABCs in their series, but only half had elevated pressures. That the cysts represent a vascular degenerative process of other more

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common lesions, such as giant cell tumors, hemangiomas, osteblastomas, and chondroblastomas, has been proposed. While this may be true for secondary lesions in which ABCs coexist with other lesions, it may not be the etiology for primary lesions. In Mankin and colleagues series [3], only 1 out of 150 primary lesions fit this description. A traumatic etiology followed by an aberrant reparative process has also been proposed but remains unsubstantiated [16].

An underlying genetic cause has been postulated, suggesting that ABCs are neoplastic in nature. Cases describing lesions in father and daughter [17] and in monozygotic twins [18], among others, have been reported. Chromosomal analysis of ABCs has shown characteristic changes with translocations in chromosome 17 [19–21] and chromosome 16 [21,22]. Oliveira and colleagues [23] described the USP6 oncogene being fused to the highly active CDH11 promoter. This occurred in spindle cells in 36 of 52 primary ABCs but was not present in the secondary ABCs analyzed. No difference in recurrence rate was noted between the lesions with or without the CDH11-USP6 fusion [23]. In combined giant cell and ABC lesions, each tumor has been shown to retain its characteristic karyotypic abnormality [24], also suggesting ABCs are a neoplastic entity. The underlying etiology of ABCs remains enigmatic and the morphologic and histologic appearance may be a common pathway for lesions occurring through different processes.

Natural history and presentation

Little is known about the natural history of the disease. It has been observed that no patient in any of the larger series died from the disease, and spontaneous regression of the lesions have been described as well. Osseous ABCs generally affect the pediatric population with an almost equal female to male ratio, with one large retrospective series (1092 patients) reporting the male to female ratio as 1.16:1 [5]. ABCs in the spine most commonly present in patients younger than 20 years of age, however, have been reported in patients ranging from 2 to 67 years [4,5].

Most series describe patients having an insidious onset of back pain with an average of 2 years' duration, but case reports have described ABCs being associated with vertebral plana [25], acute vertebral collapse (20%) [4,26], and (rarely) spinal cord injury [27–31], although these remain rare. In Boriani and colleagues series of 44 patients [4],

44% of patients had an associated axial deformity, although only 17% had clinically apparent scoliosis or kyphosis.

Imaging

Diagnosis of the condition involves plain radiographs and cross-sectional imaging with CT (Fig. 1) and contrast-enhanced MRI (Fig. 2). The radiographic and CT appearance of an ABC in the spine typically shows involvement of the posterior elements with associated vertebral body involvement [10,32–36]. A “blown-out” or “ballooned” expansile lytic lesion is apparent in the host bone on radiograph (see Fig. 1). The lesion involves a soft-tissue mass surrounded by a cortical bone that is usually delicate, described as a “shell,” and has a trabeculated appearance. Flocculent densities may be apparent within the lesion [10]. Fluid–fluid levels on CT are highly sensitive (87.5%) and specific (99.7%) [37]. The typical MRI appearance shows fluid–fluid levels [28,35], although the sign itself is not sensitive (77%) or specific (67%) [38]. For example, in a series of 700 patients, of whom 27 were diagnosed with an ABC, only 10 presented with fluid–fluid levels. Other lesions with fluid–fluid levels included osteblastoma, chondroblastoma, simple bone cyst and telangiectatic osteosarcoma, giant cell tumor, and brown tumor [39]. The presence of septa and lobulation, however, are most specific for an ABC [38].

Treatment

The treatment of an ABC of the spine depends on the region of the spine it is located, the anatomic location of the tumor, and whether there is evidence of spinal cord compression or compromise to the structural integrity of the host bone (spinal segment). A biopsy of the lesion may be undertaken if the radiographic features are not pathognomonic for ABC, for example, if the lesion is secondary or a solid variant. Fine-needle biopsy may reveal hemorrhagic tissue and may be nondiagnostic. Therefore, open biopsy is the preferred choice in conjunction with curettage following a frozen section. Histology should reveal histioblasts, giant multinucleated cells, thin capillary vessels, and hemosiderin granules [15]. In cases in which the ABC is secondary, it is essential that the biopsy be carefully planned to be consistent with oncologic principles and not



Fig. 1. CT and radiographic images of this L5 ABC demonstrate an expansile, cystic lesion with primary vertebral body involvement. (A) Axial CT. (B) Coronal CT reconstruction. (C) Sagittal CT. (D) Lateral radiograph.

compromise definitive treatment. The biopsy should be performed at a medical center familiar with the treatment of spinal malignancy and should ultimately be planned or performed by a surgeon trained in musculoskeletal oncologic principles.

Embolization

Treatment choices for spinal ABCs consist of arterial embolization, percutaneous intralesional injection of a fibrosing agent, radiotherapy, intralesional curettage with or without bone grafting,

and en bloc resection [4,40–46]. There are no prospective data on the different treatment methods, and most series describe various methods and combinations of treatments for their patients [3,4,40,47].

Embolization of ABCs has been reported with success for complete ablation of the lesion or used adjunctively to control intraoperative bleeding before curettage. Dubois and colleagues [48] reported on 17 patients in whom sclerotherapy was performed using Ethibloc (zein alcohol) and Histoacryl glue with a mean follow-up of 57 months in which there were no recurrences.

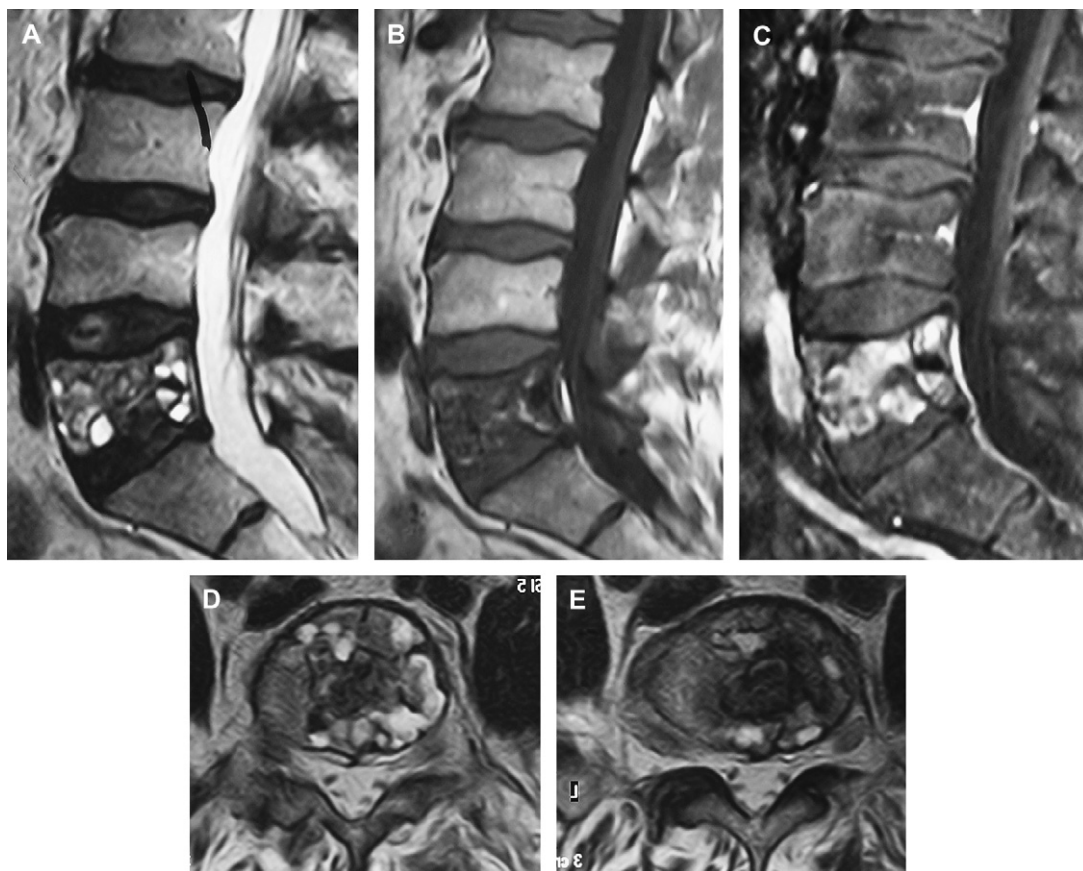


Fig. 2. MRI imaging of an L5 ABC showing a cystic lesion with heterogenous signal characteristics and heterogenous contrast enhancement. Fluid–fluid levels are also evident. (A) Sagittal T2 image. (B) Sagittal T1 image. (C) Sagittal T1 with gadolinium image. (D, E) Axial T2 images.

Each patient was treated 2 to 5 times, however. Guibaud and colleagues [42] have reported successful percutaneous embolization in 13 of 16 cases using zein alcoholic solution, although this series had only two patients who had spinal lesions. Mohit and colleagues [49] also described a successful stand-alone embolization of an ABC in the atlas in a 10-year-old girl. Gladden and colleagues [50] reported a successful yet alternative method to embolization to the upper cervical spine using used methylprednisolone and calcitonin percutaneously injected through a CT-guided approach. Despite these encouraging reports, caution is required, because neovascularity may allow the embolic material to embolize outside the lesion. Regions of concern include the cervical spine in which the vertebral arteries may be involved or the thoracolumbar junction in which the artery near artery of Adamkiewicz is present. Boriani

and colleagues described neovascular connections to the vertebral artery in a patient following recurrent cyst formation initially treated by embolization. Peraud and colleagues [45] reported a fatal embolization while treating a high cervical lesion, secondary to embolic material (Ethybloc) flowing to the vertebro-basilar system and infarcting the brainstem.

Surgery

Open surgical procedures historically have included curettage with or without bone graft and resection. In Boriani's series of 41 patients, 2 patients received en bloc resection and 32 patients received curettage, with 16 of 32 patients having only partial curettage and 14 of 32 patients receiving adjuvant embolization. Radiotherapy was used alone in three patients. Only two

patients had a recurrence, one following arterial ablation and the other following incomplete curettage. Papagelopoulos and colleagues [44] described a series of 52 patients in whom 19 of 52 had intralesional excision with bone grafting and 21 of 52 had curettage alone. Recurrence occurred within 6 months of treatment in 12 of 52 patients. In Mankin and colleagues series [3] in which 11 ABCs were located in the spine, only 1 recurred. The recurrence rate in the entire series of 150 cases was 21 of 101 for those treated with curettage and allograft, compared with 3 of 9 patients treated with curettage alone and 0 of 11 treated with en bloc resection. Ozaki and colleagues [51] had recurrence in 2 of 2 patients treated with curettage alone. de Kluver and colleagues [40] described 6 of 10 patients who had an incomplete curettage with a recurrence. The investigators further noted that complete curettage through a posterior-based approach was technically challenging, and a higher recurrence rate was associated with the posterior approach [40]. Thus, for cases with significant involvement of the vertebral body, an anterior approach may provide a more complete removal of the lesion. Garg and colleagues [52] reported 0 of 8 recurrences in which intralesional curettage, high-speed burring of the lesion, electrocautery, and bone grafting was performed, in contrast to 4 of 4 recurrences in patients who had only intralesional curettage and bone grafting. This supports the principle that complete curettage of the lining of the cyst wall is necessary to reduce the recurrence rate. Whether a lower recurrence rate occurs for patients when adjuvant embolization is used is uncertain. En bloc resection has the best chance of avoiding local recurrence. In recalcitrant recurrent lesions, radiotherapy has been used. Complications of radiotherapy have included sarcomatous degeneration of the lesion [44,53]; it is generally reserved for cases in which more conventional therapies have failed.

Intra-operative bleeding may be avoided by using selective arterial embolization, although caution has to be used around the vertebral arteries and the artery of Adamkiewicz. En bloc resection provides removal of the lesion with controlled blood loss; however, this procedure remains technically challenging and carries a high morbidity rate.

Because 50% to 80% of ABCs occur in patients younger than age 20 years and 1 of 3 ABCs can occur in the cervical spine, special attention has to be paid to post-laminectomy kyphosis. Likewise, instability in the thoracic

spine following curettage of a facet joint deserves attention. As a result, limited fusion is required to prevent a sagittal or coronal plane deformity from occurring and it should be considered during formulation of the treatment plan. Similarly, the procedure used to treat ABCs must stabilize the spine while providing for the safest and most complete resection possible.

Treatment approach

Although there has been little in the way of clinical outcome studies for ABCs, an approach to the treatment has been put forward by Boriani and colleagues [4], in 2001. They recommended the following treatments:

1. Selective arterial embolization if the diagnosis was certain, where there was no neurologic compromise and stability was not concern, and where embolization could be safely performed.
2. Complete curettage, preferably preceded by selective embolization, if pathologic fracture was a concern or in anterior lesions where there was neurologic compromise. Curettage was also recommended if embolization was not possible or for treating local recurrence.
3. En bloc excision was recommended for ABC in the posterior elements.
4. Biopsy should be performed via an open technique if the workup was not diagnostic. Curettage should be performed at that time if the biopsy was confirmatory.
5. Reconstruction should be performed if needed at the time of resection, but these authors recommended bracing for children until skeletal maturity rather than primary fusion. They recommended posterior fusion for adults if posterior arch removal resulted in the removal of one articular joint or more. If vertebral body resection is needed, anterior reconstruction must be performed.

Summary

Although there has been little in the way of clinical outcome studies for ABCs, an approach to the treatment has been put forward by Boriani and colleagues [4] in 2001. ABCs remain enigmatic with respect to their etiology, and the natural history of the disease is unknown. Although uncommon, pathologic fractures, spinal deformity, and spinal cord compression can occur as a result of these

lesions. Diagnostic difficulty can occur with secondary ABCs and the rare solid variants. If the diagnosis is in question, an open biopsy followed by curettage should be performed. Curettage must involve complete removal of the lining of the cyst and this may induce significant intraoperative bleeding. An anterior approach may provide a more complete removal of the lesion. Intraoperative bleeding may be avoided by using selective arterial embolization, although caution has to be used around the vertebral arteries and the artery of Adamkiewicz. Embolization, however, has been used effectively in difficult to treat regions, such as the high cervical spine. En bloc resection provides removal of the lesion with controlled blood loss; however, this procedure remains technically challenging and carries a high morbidity rate.

Large lesions in the spine are associated with pain and occasionally may underlie lead to kyphosis or scoliosis. As the lesions enlarge, spinal instability may occur. Treatment must be individualized, and patients should be counseled about recurrence of the lesion despite the method of treatment.

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