Case Report

Pelvic Aneurysmal Bone Cyst: Case report and Literature Review

Mohammed Al Sobeai¹, Wazzan Al Juhani²

Abstract

**Introduction:** An aneurysmal bone cyst (ABC) is an enigmatic lesion that can involve any bone. Pelvic ABC is a challenging lesion that is sometimes difficult to manage surgically.

**Case Report:** We present a case of ABC involving the right ilium and supra-acetabular area. Surgical resection was performed after embolization, to reduce intraoperative bleeding, and was followed by acetabular reconstruction.

**Conclusion:** Pelvic ABC is a challenging tumor. Management depend on size of the lesion and stability in addition to clinical presentation. Small lesions can be treated conservatively. Large lesions affecting pelvic mechanical stability should be treated with surgical resection and reconstruction.

**Keywords:** Aneurysmal bone cyst, Pelvis, benign tumor.

**Introduction**
The ABC lesion has been reported in the orthopedic literature for more than 60 years; it was first described in 1942 by Jaffe and Lichtenstein[1]. It represents almost 1% of primary bone lesions[2-4]. ABCs are primary lesions in 70% of cases and in 30% arise from an eosinophilic granuloma, a simple bone cyst, a chondroblastoma, a giant-cell tumor (GCT), or an osteosarcoma[5-8]. Pelvic ABCs are intractable tumors because they may affect acetabular or sacral stability and are usually challenging surgically.

**Case Report**
A 24-year-old female presented to the orthopedic oncology clinic with right hip pain associated with a mass. The mass was large but had been growing slowly over 6 months. The patient has no referred pain and was still walking. On examination, she had a large mass fixed to her pelvis over the right side (Fig. 1). Plain radiography showed a large lytic lesion involving the right innominate bone with ill-defined margins in some areas and extending to the right acetabular dome (Fig. 2). A pelvis MR1 (Fig. 3A, B) showed multiple fluid-fluid levels, indicating primary versus secondary ABC. A systemic investigation to rule out distant disease was normal.

A biopsy was performed initially percutaneously with a core needle. Our pathologist could not identify any malignant cells and saw mainly hemorrhagic cysts. Then, the patient underwent an open biopsy to provide more tissue. The pathologist confirmed the diagnosis of primary ABC. After diagnosis, surgery was planned. Embolization was performed 24 h before resection. In the operating room, a skin incision was created from the middle of the iliac crest to the anterior superior iliac spine (ASIS) and down to the pubic rami (Fig. 1). Another line extended from ASIS to the greater trochanter. Muscles were dissected around the mass from the outer to the inner side of the ilium. The neurovascular bundle was identified and protected at the femoral canal (Fig. 4). The mass was too large to be removed en bloc and was released in pieces (Fig. 5). Part of the mass extended to the dome of acetabulum, which was reconstructed with

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The pathophysiology of ABC remains unclear. The name is merely descriptive and no genetic or neoplastic cause has been identified[10]. Many theories have been proposed, including vascular, traumatic, and genetic etiologies. Campanachi and Lichtenstein believed in vascular theory and described the pathophysiology as a local vascular disturbance and hemorrhage[11-14]. Ratcliffe and Grimmer described the formation of ABC at the proximal metaphysis of a tibia fracture and considered that ABC occurred secondarily to vascular disturbances initiated by a fracture[15]. A third theory of ABC pathophysiology is the genetic theory. The genetic theory indicates that primary ABC is neoplastic rather than reactive. Oliveira reported a genetic translocation, t(16,17) which was absent in secondary ABC[16]. Extensive studies of the role of insulin-like growth factor-I in bone tissue have suggested that it plays a role in the pathogenesis of GCT and ABC[17]. Familial cases of ABC have been reported, supporting the genetic and neoplastic theories[11, 18-24]. Malignant transformation has been reported, mainly after radiotherapy of an ABC or initially misdiagnosed telangiectatic osteosarcoma. However, few cases have developed into malignancies without a history of previous radiotherapy[11, 21, 25, 26]. Histological examinations of ABC reveal multiple cavities and of various sizes, filled with blood and/or proteinaceous material.

These cavities are lined with endothelial and smooth muscle cells. The septa consist of several cell types, including spindle cells, inflammatory cells, and some giant cells. Osteoid formation, which is seen in ABC, is not seen in GCT tumors. 5 – 10 % of ABC are atypically solid in nature with little or no cystic formation[10, 11, 13,14, 27].

**Literature Review**

Review of literature revealed 25 articles discussed the pelvic aneurysmal bone cyst. Articles that reported different body parts including the pelvis were excluded. 25 reports and series were identified. Around 59 males and 55 females are reported with different parts of the pelvis involved. Age was ranging from 18 months to 64 years old. Management was studied in all cases and discussed below.

**Clinical Presentation**

ABC can occur at ages from 1 to 59 years, with a peak at 12-13 years. It also affects males more than females. Usually, ABC presents with pain and swelling secondary to the expansile mass. Secondary symptoms, such as fever, weight loss, nausea, and vomiting, that typically occur with malignancies are not common[10]. Pelvic ABC has several unique characteristics, including proximity of the tumor to the neurovascular bundle and acetabulum, making it more difficult to approach and reconstruct, intraoperative bleeding, and that pelvic ABC tend to be large and highly vascular[11, 28-30]. Usually, they present with pain in the low back, groin, or thigh, commonly associated with a limp. Sacral lesions can present with radicular symptoms and may be similar to spinal disc disease due to encasement of the cauda equina or nerve roots[11, 30, 31].

**Imaging**

**Figure 1.** huge tumor over the right side of the pelvis. Picture is taken immediately before surgery.

A bone graft, calcium carbonate based. A drain was placed and closure was achieved in layers. Histopathology slides were shown in (Figs 6 and 7). Post-operative radiography taken on the first day postoperatively is shown in (Fig 8). The patient started to mobilize with full weight-bearing the next day and was discharged to home after a few days with no postoperative complications. She was seen in the clinic at 2 weeks for a wound check and removal of sutures and then at 6 weeks, 3 months, and 6 months. Radiographs are shown in (Fig 9). The last clinical visit was at one year postoperatively and the patient is mobilizing well with no hip or pelvic pain.

**Discussion**

This case is a rare condition where large lesion in the pelvis where stability is affected and needed reconstruction with bone graft. The lesion was large and involving the acetabulum. Mechanical stability was an issue and reconstruction with bone graft was done. We have chosen this approach as literature was supporting the technique in similar lesions involving the acetabulum.

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**Imaging**

**Figure 3a and 3b:** - MRI of the pelvis including (A) axial and (B) sagittal views.

**Figure 2.** Preoperative radiography.

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Figure 4. Femoral neurovascular bundle was identified and protected during the approach.

Figure 5. Tumor after resection.
Imaging of pelvic ABC is important in diagnosis[32]. Combining conventional radiography with MRI increases the sensitivity and specificity of the diagnosis[32]. Similar lesions that can be misdiagnosed are GCTs and telangiectatic osteosarcoma. Typically, ABC is seen on plain radiographs as an eccentric expansile lesion involving the metaphysis. Usually, it is contained within a thin shell of cortex. Thick nodular enhancement at the rim of the tissue on MRI and CT is suggestive of telangiectatic osteosarcoma rather than ABC. These nodularities are secondary to osteoid mineralization, due to sarcomatous cells[33].

Epiphyseal ABC should raise the suspicion of a lesion secondary to benign or malignant lesions. However, there are few reported cases of epiphyseal primary ABC in the literature[11, 34, 35]. Dabska and Buraczewski described four radiological phases of the disease (Table 1)[10]. Computed tomography and MRI usually show multiple fluid levels in multiloculated lesion. This is not a pathognomonic sign of ABC, because it may also be seen in telangiectatic osteosarcoma, GCT, chondroblastoma, secondary ABC, fracture through a simple cyst, and even conventional osteosarcoma post-chemotherapy[36].

**Treatment**

Spontaneous resolution of ABC may occur through thrombosis and fibrosis after no or partial treatment. Malghem et al. reported three cases with spontaneous resolution and suggested conservative management of ABC when there is no risk of fracture or stability issues[11, 37, 38].

**Embolization**

Embolization of pelvic ABC is recommended before curettage and bone graft, especially if the ABC is large. Extensive intraoperative bleeding may occur if the tumor is opened, due to loss of the tamponade effect of a closed compartment[39]. However, some believe that ABC can be treated effectively without surgery. Rossi et al. reported a success rate of 94% without surgery. The need to repeat embolization was more frequent in patients younger than 16 years old and with lesions of more than 5 cm in diameter[40].

**Bisphosphonates**

One case of a sacral ABC was reported by Simm in 2013 after a failed embolization because no supplying vessel was identified. The patient was then administered zoledronic acid at 0.04 mg/kg. After seven injections at 4-month intervals, the tumor resolved, as confirmed by MRI[41].

**Sclerotherapy**

Polidocanol shows excellent results with ABCs. Additionally, it is safe and causes only transient inflammatory reactions when it leaks into surrounding tissues. Brosjo et al. found that it was effective in ABC, especially ABC of the pelvis. All six pelvic and sacral ABCs treated with sclerotherapy healed without complications[42]. Varshney et al. reported a disturbance of growth in 4% of cases treated with polidocanol. However, only 21% healed after the first injection. The remaining patients required repeated injections[43]. Ehibloc has also been used in the treatment of ABC. However, this agent has been abandoned because of the many complications reported[11, 19, 44-48].

**Steroid and bone graft injections**

Steroid injections are not common practice in ABC. Unlike the unicameral bone cyst, Scaglietti reported that ABC responded poorly to steroid injections; all 12 of their cases then underwent curettage and bone graft[49]. Bone graft is a minimally invasive procedure performed through a small incision. Demineralized bone matrix (DBM) mixed with bone marrow is injected into the lesion. It stimulates healing of the lesion without removal of the lesion contents. Docquier and colleagues published their experience with DBM in 2005[50].

**Curettage and bone graft versus en bloc resection**

Treatment strategies depend on the size of the lesion and the part of the pelvis and sacrum involved. Lesions < 5 cm can be treated with curettage and bone graft and rarely require reconstruction. However, lesions > 5 cm that involve the cortex and affect the integrity of acetabulum and sacrum require more aggressive treatment:
that is, excision and reconstruction. A structural autograft or allograft can be used to rebuild the structural integrity[51].

Adjuvant therapy
The available adjuvant therapies can reduce recurrence and improve outcome. Phenol is well known but cannot be used in sacral lesions near nerve roots because it may damage the nerves[30] Marceco and Peeters reported good experience in cryosurgery with liquid nitrogen[11, 52, 53]. Combining cryosurgery and curettage will reduce the need for major reconstruction[54]. Adjuvant radiotherapy is not an option for ABC, except in cases where surgery is not possible. Post-operative sarcoma is a potentially devastating complication that prevents the use of this option[42]. Other possible complications include growth arrest with secondary deformities and gonadal arrest[55, 56].

Complications
Pelvic ABC may be complicated with pathological fractures that make reconstruction challenging. Acetabular fracture, protrusion acetabuli, destruction of the sacroiliac joint, and sacral nerve compression are the possible complications[28].

Conclusion
Pelvic ABC is a challenging tumor because of its location. An appropriate approach is essential to confirm the diagnosis and to rule out other malignant conditions that will prevent problems in management. Conservative management can be offered for small lesions and those that do not affect stability. In large lesions or where stability is an issue, surgery is necessary and reconstruction will facilitate resumption of normal functioning.

Clinical Message
Pelvic ABC is a challenging tumor because of its location. An appropriate diagnostic approach is essential to confirm the diagnosis and to rule out other malignant conditions. Conservative management options can be offered for small lesions and those that do not affect stability. In large lesions or where stability is an issue, surgery is necessary and reconstruction will facilitate resumption of normal functioning.

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