

Aneurysmal Bone Cyst of the Pelvis: A Challenge in Treatment: Review of the Literature

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Citation

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Abstract

Aneurysmal bone cysts are amongst the common entities which can affect any bone of the body. Among them, though, aneurysmal bone cysts of the pelvis are not so uncommon. It is a challenging job to deal in respect to its treatment. Therefore we reviewed the literature regarding aneurysmal bone cysts of the pelvis and its treatment modalities in context with our experience in treating aneurysmal bone cysts of the pelvis at out hospital.

INTRODUCTION

Aneurysmal bone cysts, first described by Jaffe and Lichtenstein in 1942 is non-neoplastic expansile lesion consisting of blood filled spaces separated by connective tissue septa containing bone or osteoid and osteoclast giant cells. The etiology is unknown. These may be primary or secondary. ABC is an uncommon expansile osteolytic lesion of bone consisting of a proliferation of vascular tissue that forms a lining around blood filled cystic lesion; It develops in metaphyseal region of long bones, pelvis, vertebral posterior elements. It commonly involves the proximal humerus, femur, tibia, and pelvis. It can cause paraplegia when it involves the vertebral posterior elements. Associated or adjacent Lesions may be a secondary ABC in 30% of cases GCT; chondroblastoma;osteoblastoma;osteosarcoma.

They may easily be mistaken for a malignant tumor both radiographically and pathologically. These diagnostic problems are due to their rapid growth, extensive destruction of bone, wide extraosseous tumor masses, and marked cellular exuberance. The differential diagnosis of aneurysmal bone cysts including giant cell tumor, calcified solitary bone cysts, low-grade osteosarcoma, and telangiectatic osteosarcoma becomes even more complicated when the lesion arises at sites other than the long bones and presents with extensive extraosseous, soft-tissue tumor masses. The latter cases—especially when they occur as sacral or presacral tumors—present challenges with respect to successful treatment, which should combine surgical removal of the entire lesion following oncological criteria to prevent recurrences and osteosynthesis to

guarantee the biomechanical stability of the spinal-pelvic junction.¹ Here we report on the clinical case of a female patient with an aneurysmal bone cyst of the iliac bone. The report includes the diagnostic challenges, the surgical options, the histopathological findings, and long-term clinical and radiographic follow up.

EPIDEMIOLOGY

- peak incidence in 2nd decade
- 80% by age 20
- male : female is 1:1.3
- incidence is 0.14 / 100,000
- 1% of bone tumors

NATURAL HISTORY

- variable Growth Rate
- indolent lesions may spontaneously involute (months to years)
- spontaneous resolution very uncommon in aggressive or secondary lesions
- no documented cases of malignant transformation (telangiectatic osteosarcoma)
- Pathophysiology

Trauma is considered an initiating factor in the pathogenesis

of some cysts in well-documented cases involving acute fracture. Local hemodynamic alterations related to venous obstruction or arteriovenous fistulae that occur after an injury are important in the pathogenesis of an aneurysmal bone cyst.

The lesion is a component of, or arises within, a preexisting bone tumor in about one third of cases; this finding further substantiates the fact that aneurysmal bone cysts occur in an abnormal bone as a result of associated hemodynamic changes. An aneurysmal bone cyst can arise from a preexisting chondroblastoma, a chondromyxoid fibroma, an osteoblastoma, a giant cell tumor, or fibrous dysplasia. Less frequently, it results from some malignant tumors, such as osteosarcoma, chondrosarcoma, and hemangioendothelioma.

Aneurysmal bone cysts may be purely intraosseous, arising from the bone marrow cavity. In this case, they are primarily cystic and slowly expand into the cortex. They may be extraosseous, arising from surface of bones, eroding adjacent cortex, and extending into the marrow space.

Four phases of pathogenesis are recognized, as follows:

- Osteolytic initial phase
- Active growth phase, which is characterized by rapid destruction of bone and a subperiosteal blow-out pattern
- Mature stage, also known as stage of stabilization, which is manifested by formation of a distinct peripheral bony shell and internal bony septae and trabeculae that produce the classic soap-bubble appearance.
- Healing phase with progressive calcification and ossification of the cyst and its eventual transformation into a dense bony mass with an irregular structure.

RACE: No specific racial distribution has been identified.

SEX: Compared with males, females have an increased incidence.

AGE: Aneurysmal bone cysts may occur in patients aged 10-30 years, with a peak incidence in those aged 16 years. About 75% of patients are younger than 20 years.

ANATOMY: Regarding the location of the lesions, any bone may be affected. Approximate frequencies by site are shown

below:

- Skull and mandible (4%)
- Spine (16%)
- Clavicle and ribs (5%)
- Upper extremity (21%)
- Pelvis and sacrum (12%)
- Femur (13%)
- Lower leg (24%)
- Foot (3%)

The most common site is the metaphyseal region of the knee.

Short tubular bones are less frequently affected and are involved in about 10% of cases.

Spinal involvement demonstrates a predilection for the posterior elements. In decreasing order of frequency, the following parts of the spine are involved: cervical, thoracic, lumbar. Contiguous vertebrae may be involved in 25% of cases.

The cyst involves the diaphysis in isolation in about 8% of cases.

INVESTIGATIONS

RADIOGRAPHS

- radiolucent lesion with expanded cortex arising in medullary canal of metaphysis;
- aneurysmal expanded appearance of cortex is contained by periosteum & thin shell of bone;
 - marked cortical thinning and erosion and periosteal elevation;
 - this lesion rarely penetrates the articular surface or growth plate;
 - radiographic differential diagnosis includes:
 - simple bone cyst
 - central location (unlike ABC)
 - absence of expansion

- lack of cortical discontinuity
- giant cell tumor of bone
 - lack of expansion
 - begin in epiphysis with extension into metaphysis
 - more likely to be centrally located
- telangiectatic osteosarcoma
 - difficult to distinguish radiographically from an aggressive ABC
 - angiosarcoma.
 - osteoblastoma
 - may have a "soap bubble" expansile appearance
 - no fluid level on CT/MR
 - CT scan:
 - look for fluid - fluid level (blood / serum): fluid-fluid level may also be seen in telangiectatic osteosarcoma;
 - ABC has a density of about 20 hounsfield units;
 - Bone Scan:
 - shows intense uptake in the margin of the lesion, with normal background or decreased uptake in its center;
 - MRI:
 - Bright on T2 and fat suppression,

intermediate on T1

DISCUSSION

McQueen et al. described two cases of aneurysmal bone cyst in the pelvis which healed spontaneously are described. Spontaneous regression of aneurysmal bone cysts has not previously been well documented. Our experience with these two cases suggests that radical intervention is not always necessary.²

Capanna reviewed twenty-three cases of pelvic aneurysmal bone cysts treated at the Istituto Ortopedico Rizzoli after a mean follow-up of 7 years. Eighteen cysts involved the anterior arch, four extended into the iliac wing and the anterior arch, and one invaded the entire hemipelvis. The acetabulum was involved in 56.5% of the cases. Fourteen patients were treated with surgery (curettage 11; resection 3), and five with radiation therapy; two patients had both modalities; two additional patients refused any treatment after biopsy. The overall recurrence rate was 13% (one case after curettage, one after radiation therapy, and one after combined treatment). Significant complications affected the final functional result in four of seven patients who received radiation therapy, while only one minor complication was seen in the surgical group.³

On the assumption that an aneurysmal bone cyst has an intrinsic potential to heal by ossification, a new, minimally invasive protocol was developed. Docquier et al. introduced demineralized bone powder mixed with bone-marrow aspirate into the cyst to halt the expansion phase and to allow the cyst to ossify. They hypothesized that, in order to induce bone-healing, cells from the cyst are needed to respond to the inductive material but that curettage or extensive surgery is not necessary. The goals of their study were to assess cyst-healing and to determine the prevalence of recurrence associated with this new procedure. Thirteen biopsy-proven primary aneurysmal bone cysts were entered through a small incision, and a paste of demineralized bone and autologous bone marrow was introduced with an applicator. The study group included three male and ten female patients with a mean age of 16.6 years. The cyst was located in a long bone in six patients, the pelvis in five patients, and the scapular glenoid and the calcaneus in one patient each. Five patients had not received treatment previously, whereas one had had a preoperative embolization and seven had recurrent lesions that had been treated previously. After a mean duration of follow-up of 3.9 years, healing was achieved in eleven patients. They concluded that

this minimally invasive method is able to promote the self-healing of a primary aneurysmal bone cyst. As no curettage is required, the proposed treatment avoids extensive surgery and blood loss and is convenient for the treatment of poorly accessible lesions such as those occurring in the pelvis.⁴

Cisneros et al. studied the treatment of a patient with an aneurysmal bone cyst involving the pelvis by selective arterial embolism. After one year of study, pain relief was complete and radiologically the lesion was healing with increased density of the cyst wall and intracystic trabecular bone formation.⁵

Papagelopoulos et al. studied forty consecutive patients with an aneurysmal bone cyst of the pelvis and/or sacrum which were treated from 1921 to 1996. Their medical records and radiographic and imaging studies were reviewed, and histological sections from the cysts were examined. Seventeen lesions were iliosacral, sixteen were acetabular, and seven were ischiopubic. Seven involved the hip joint, and two involved the sacroiliac joint. All twelve sacral lesions extended to more than one sacral segment and were associated with neurological signs and symptoms. Destructive acetabular lesions were associated with pathological fracture in five patients and with medial migration of the femoral head, hip subluxation, and hip dislocation in one patient each. The mean duration of follow-up was thirteen years (range, three to fifty-three years). Thirty-five patients who were initially treated for a primary lesion had surgical treatment (twenty-one had excision-curettage and fourteen had intralesional excision); two patients also had adjuvant radiation therapy. Of the thirty-five patients, five (14%) had a local recurrence noted less than eighteen months after the operation. Of five patients initially treated for a recurrent lesion, one had a local recurrence. At the latest follow-up examination, all forty patients were disease-free and twenty-eight (70%) were asymptomatic. There were two deep infections. They concluded that aneurysmal bone cysts of the pelvis and sacrum are usually aggressive lesions associated with substantial bone destruction, pathological fractures, and local recurrence. Current management recommendations include preoperative selective arterial embolization, excision-curettage, and bone-grafting.⁶

We studied 10 patients of Aneurysmal Bone of pelvic presented to Department of Orthopaedics, B P Koirala Institute of Health Sciences, Dharan, Nepal. Most of the patients presented with dull boring type of pain, and

progressively increasing swelling on the groin, and with only progressive swelling on the loin region. Most of the patients had no history of trauma and any constitutional symptoms like fever, cough, chest pain, altered bowel habits, multiple joint pains etc. On examination, there were ill defined swellings with definite tenderness. There were not inguinal lymphadenopathy and no abnormal bruit heard over the swellings. On plain radiographs, an expansile radiolucent lesion with soap bubble appearance present on the pelvic bone, 5 in iliac bone, 2 in ischium, 2 in iliopectineal, 1 in pubic rami. A radiographs of a patient with ABC pelvis on the iliac bone without breach in cortex with preservation of the superolateral roof of acetabulum was shown in Fig A. CT scan showed well circumscribed radiolucent lesion on the iliac bone without cortical breach (Fig B). In all cases histopathological examination was done to confirm Aneurysmal Bone cyst of pelvis (Fig C).

Figure 1

Figure A: showing ABC of Right Iliac bone of pelvis

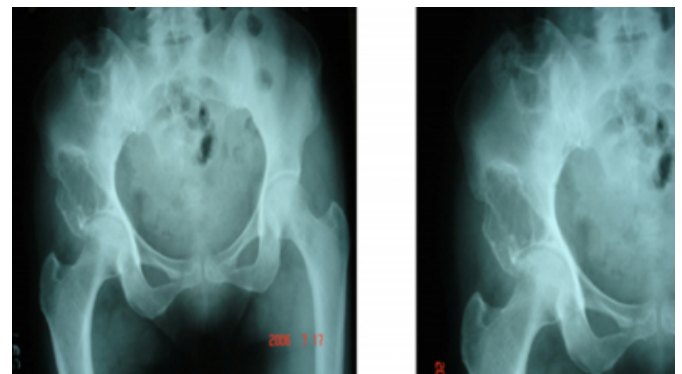


Figure 2

Figure B: showing transverse cut section of ABC iliac bone in CT scan

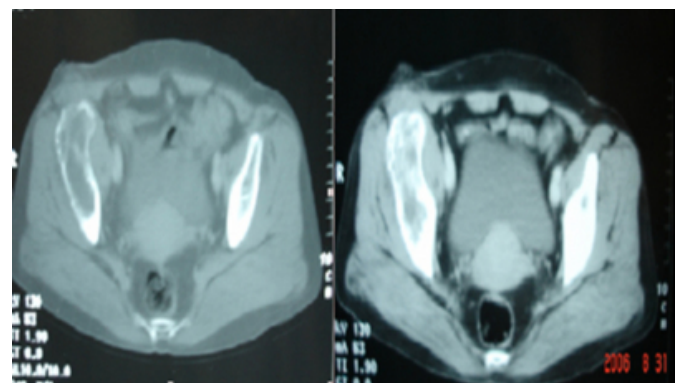
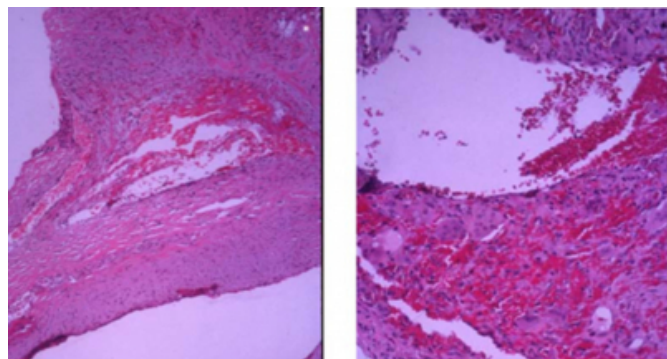


Figure 3

Figure C: showing histopathological picture of Aneurysmal bone cyst



Aneurysmal bone cysts of iliac bone with expansile lesions were approached through Smith Peterson. Thorough curettage of the lesions was done; hemostasis was achieved with the help of bone wax and Jelfoam. The cavity was filled with cortical cancellous graft harvested from opposite iliac crest and fibula along Z-bone. During the procedure, we were cautious about not damaging the roof of acetabulum. The perioperative and post operative period was uneventful. The patients were advised non weight bearing crutch walking for 3 months, after which kept on partial weight bearing crutch walking. After the follow up of 6 months, patients were allowed to bear weight without support after evaluating radiological consolidation of the cyst. At one year all patients had no complains at all.

CONCLUSION

Pre operative selective arterial embolization is one of the ideal steps in management of aneurysmal bone cysts to control bleeding prior to curettage and bone grafting. But, in our context, it is very sophisticated measure and can not be

affordable. As per literature, aneurysmal bone cysts of the pelvis were treated without intervention that is by spontaneous healing and some treated with only selective arterial embolization. Till date, excision and curettage followed by autologous bone grafting is a popular treatment for pelvic aneurysmal bone cysts, therefore it was adopted in our cases, which had very good outcome in subsequent follow up and was satisfied result at one year follow up.

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