

Reconstruction of Pelvic Ring after Hemipelvectomy and Fibular graft at 39 – years – old man with Aneurysmal Bone Cyst at Iliac bone

Budi M.N.S.¹, Ismono D², Primana, H.R.^{3*}, Tua, S.J.R.³

¹ Staff of Oncology division of Ortopaedic and Traumatology Department Hasan Sadikin General Hospital, Bandung.

² Staff of Oncology division of Ortopaedic and Traumatology Department Hasan Sadikin General Hospital, Bandung.

³ Resident of Ortopaedics and Traumatology Department Hasan Sadikin General Hospital, Bandung.

*Corresponding Author E-mail: rplhendy@gmail.com

Abstract

Introduction: The ABC (Aneurysmal Bone Cysts) lesion has been reported in the orthopedic literature for more than 60 years; it was first described in 1942 by Jaffe and Lichtenstein. Aneurysmal bone cyst (ABC) in pelvic is a benign, expansile osteolytic lesion that typically affects the metaphysis of long bones in the second decade of life and occurring in patients less than 20 years. The Several ABC treatment modalities had been utilized including wide resection, intralesional resection/curettage with or without different adjuvants, radiation, embolization, intralesional sclerotherapy.

Case Report: We present a rare case of a huge aneurysmal bone cyst (ABC) in the iliac bone, occurring in the male patient 4th decade of life. The patient presented with a history of pain and huge pelvic mass size 8x6x5 cm extend to abdominal region for 5 years. Plain radiograph and computed tomography showed expansile lytic lesion arising from the all right iliac bone extend to Lumbar 5. A biopsy was performed and histology confirmed diagnosis of aneurysmal bone cyst. The patient was managed surgically with hemipelvectomy type 1 and Autogenous bone graft from fibula and reconstruction with plate and screw for pelvic ring with and re-attach the muscle on it. Follow up 5 months after surgery the patient can squat and stand maximally with no pain, nor recurrence and can return to normal activity.

Conclusion: Early diagnosis and complete resection of tumor and treatment of relevant symptoms represent a viable treatment for this rare disorder to achieve increased life expectancy, low recurrence of tumor and improved quality of life.

Keywords: aneurysmal bone cyst, pelvis, hemipelvectomy

1. Introduction

The ABC (Aneurysmal Bone Cysts) lesion has been reported in the orthopedic literature for more than 60 years; it was first described in 1942 by Jaffe and Lichtenstein¹. According to the World Health Organization Classification of Tumors, the aneurysmal bone cyst (ABC) is a benign expansile tumor like lesion of bone consisting of blood-filled cavities. The cavity space are separated by fibrous connective tissue septa containing reactive woven bone, osteoid tissue, and osteoclast type giant cells^{2,3}. They usually manifest a varying potential to be locally aggressive⁴. The incidence is recorded often affected in the second decade of life and occurring in patients less than 20 years^{5,6,7}, Of all aneurysmal bone cysts, about 8-12% occurs in the pelvis, ABCs commonly appear in the metaphysis of a long bones including femur, tibia, fibula, humerus, skull, and posterior elements of the spine.^{8,9}, the several ABC addressing the patients symptoms and prevention/treatment of fracture. The treatment modalities had been utilized including wide resection, intralesional resection / curettage with or without bone grafting, different adjuvants, radiation, embolization, intralesional sclerotherapy⁹

2. Case Report

A 39-year-old male presented to the orthopedic oncology outpatient clinic with chief complaint a right hip pain and a large mass at right pelvic. It enlarged slowly from the size of a chicken egg to size of a ball over 5 months (Figure 1). The pain is increasing and getting worse, and more pain is felt at night. Patient still can stand and walked. On Physical examination, he had a large mass fixed to the right side of his pelvis. (Figure 1). There was no history of fever, any chronic illness or swellings in other body regions. In the physical examination showed an

approximately 8x6x5 cm mass over her right iliac region extend to abdominal region. the lump was hard in consistency, well defined, irregular border, and isolated which was non-movable, warmer than the surrounding, tender on deep palpation, and crepitations were felt over the most prominent part. Movements and power of right hip were normal. (Figure 1).



Figure 1. Clinical Presentation of the Mass at right iliac wing

From the radiological examination, in a plain radiograph of pelvic showed there were expansile osteolytic lesions involving the right ilium. the entire iliac bone from the crest to the superior border of the acetabulum and to the Lumbar 5 with thin sclerotic margins (Figure 2).



Figure 2. Pelvic AP X-ray

From Computed tomography (CT) scan showed a large honeycomb type lesion of the right iliac bone destruction extend to lumbar 5, marked by expansion with a thinned cortex, and without calcified matrix. (Figure 3).

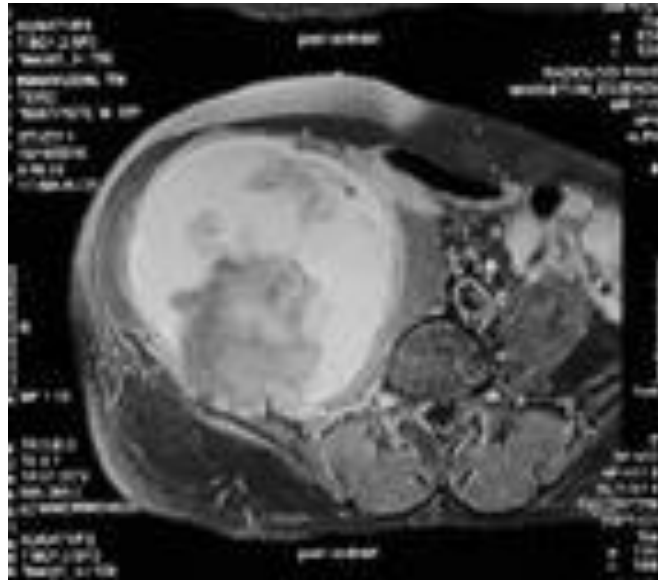


Figure 3. CT Scan axial view Shows tumor was protruded to the abdominal region

We performed a histopathology examination with an open biopsy and confirmed the lesion to be an Aneurysmal bone cyst (Fig.4). Accordance with this result, we, therefore planned to performed hemipelvectomy type I followed by Auto bone graft and reconstruction of the pelvic we performed an internal fixation with plate and screw. In the operating room, the patient was positioned in a lateral decubitus position. The skin incision from pubic rami superior through the iliac crest extends to the posterior superior iliac spine. An ellipse incision was made 4 cm margin of skin around the biopsy track along with the skin incision (Fig.4a). After the tumor mass was exposed and resect it (Fig.4b, c), and found tumor size 18x15x10 cm (Fig.5a)

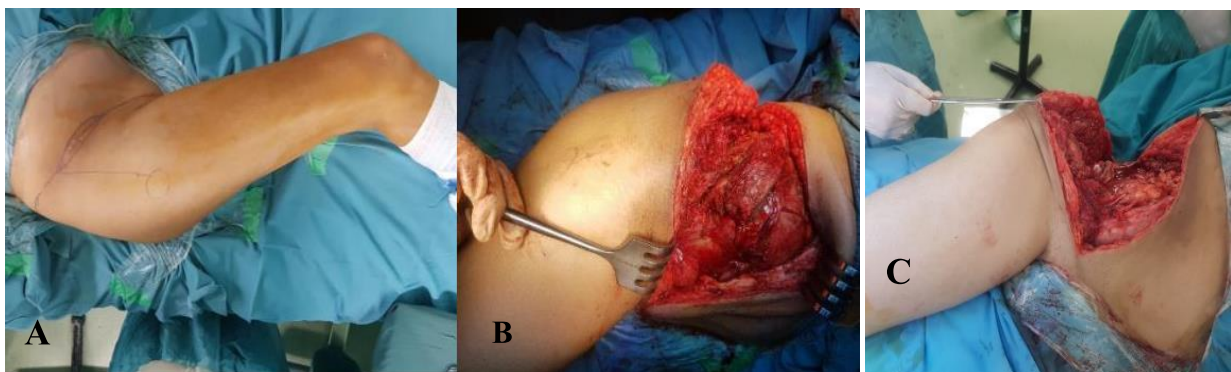


Figure 4. a) Clinical picture b) Tumor at iliac wing c) After tumor resection

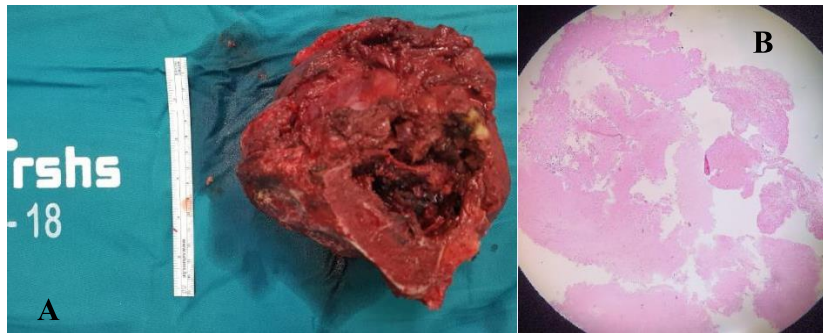


Figure 5. a) Tumor after resection size 18x15x10 cm b) Histology

After the tumor resection, we do pelvic reconstruction with autograft from the fibular bone (Fig.6a) and we did a fixation with a 10-hole reconstruction plate at the defect in the iliac wing (Fig.6b,c). Then we closed the wound and the tumor mass was sent to the Pathology Anatomy for histopathology examination diagnosis.

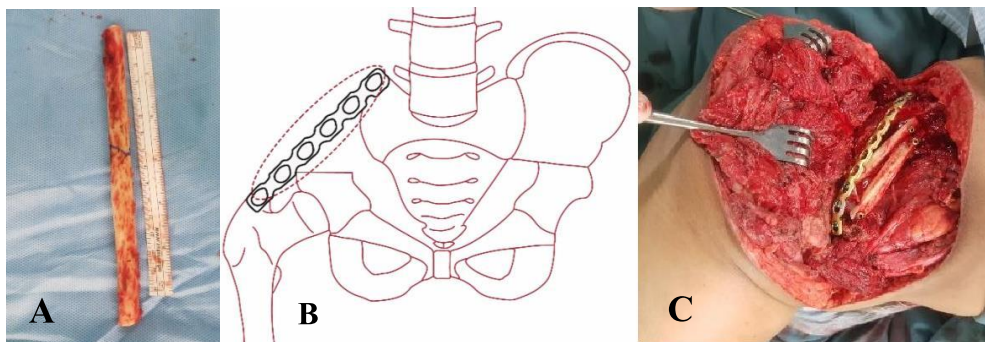


Figure 6 a) Fibular bone graft, b,c) After reconstruction

Follow up one year postoperative, the patient can do fully weight bearing, squat, normal gait, no pain, and do a normal activity living (Figure 6a).





Figure 7 a) Patient can full weight bearing and squat b) Xray Post Op

3. Discussion

The presented case has manifested atypical characteristics, such as relatively older age, less usual site (the iliac bone), and the huge size of the lesion, 11×15×10 cm. ABCs usually involve the long bones of the extremities, membranous bones of the thorax, or vertebrae. In our case, the ABC tumor involves iliac bone. In a population-based analysis of 110 surgically treated ABC patients listed in the Vienna Bone Tumor Registry, 73 ABCs (66%) occurred in patients younger than age 20 years⁹.

Clear predilection was shown in ABC cases, with only 8 – 12% of all cases occur in the pelvis. Cases of ABC occur prevalently in long bones, in sites such as femur, tibia, humerus, and fibula (accounting approximately for 52% of all sites for ABC)⁵. In cases of large ABC (exceeding 5 cm), destruction of the sacroiliac joints and acetabulum may occur.[8] The cases are most often found in young patients, aged approximately 20 years old. In a study by Mohaidat et al, most of the patients were either younger than 10 years of age or older than 20 years; with the oldest patient was 32 years old. There were only 6 of 25 patients were older than 20 years; compared to almost half of the patients were 10 years or younger⁷. The case is considered unusual due to several characteristics of the patient: older age (39 years old) and its occurrence on pelvis.

Exact pathogenesis of ABC is still unknown. Cytogenetic and molecular studies suggested neoplastic nature of ABC had noted genetic mutations, creating fusion gene. The fused genes inhibit osteoblast maturation and causes dysregulation of signaling pathways of bone

morphogenetic protein. The recent, most popular theory, regarding the pathogenesis of ABC is the involvement of capillaries on the affected bone. Increased capillary pressure in prelesion site may lead to increased blood extravasation, leading to cortical destruction. Cystic spaces on such cases are connected with the patients' capillary network; as shown by studies noting the lack of venous blood components, such as lack of venular blood clotting and confirmation by venographic studies⁹.

Diagnostic modalities for ABC often utilize imaging techniques: CT and MRI scans often provide sufficient information regarding the location and size of the cyst. Cases of ABC often present as soap-bubble like appearance in CT scans due to ballooning, multi-locular lytic lesions. Thinning of cortical wall of the lesion was brought by erosion and expansion of cortex. In cases of vertebral body collapse, pathologic partial or complete fracture may occur, particularly in ABC cases found in vertebral body. Additionally, MRI scans are useful in assessing possible soft tissue or neural involvement, particularly in cases presenting with multilocular cysts with fluid-fluid interfaces on T2-weighted images⁶. Confirmatory diagnosis for ABC requires histopathological examination of the biopsy sample from the suspected tumor. Hemorrhagic tissue and cavitation spaces (separated with fibrous septa) may be found. Spindle cells, inflammatory cells, and smaller percentage of giant cells may be found under microscope examination. Osteoid formation with or without osteoblastic rimming may be noted. A small portion of ABC cases, approximately in 5 – 10% of cases, found no cystic formation⁵. Preceding history and physical examination had noted visible tumors on pelvic region. Additional imaging studies were performed: CT scan had found a large honeycomb lesion of the right iliac bone distraction (extending to the 5th lumbar vertebra) with thinning of the cortex. Open biopsy in this case as performed to confirm the diagnosis. Following the diagnosis, en bloc resection was performed for the patient.

Complete surgical excision is recommended in treatment of ABC due to its excellent local control rates. Several other studies had noted its efficacy in preventing recurrence. The downside of the procedure is its higher rate of complications compared to other methods available for resection of the tumor. The higher rate of complication, however, was not significant. Intralesional surgical procedures may be utilized to remove the active component of the cyst and to partially disrupt blood flow to the lesion. Intralesional excisions are simpler compared to complete surgical excision while maintaining an acceptable rate of local control (approximately 90%). Minimally invasive techniques may be utilized, particularly by using

curopsey (limited curettage during biopsy by removing the lining membrane form various areas of the lesion). Approximate rate of local control was 81% with significantly shorter recovery time⁴. The patient had presented with a favorable outcome; after a period of physical therapy, the patient gained full mobility and was able to bear load (by squatting) on his pelvis. Despite the low risk of recurrence, additional follow-up may be warranted.

Surgery with hemipelvectomy, electric cauterization, and Non-vascular fibular bone graft were performed. Fixation with plate and screw were performed through the window of the iliac crest. Follow up one year postoperative, the patient can do fully weight bearing, squat and do a normal activity living. But a careful follow-up is needed to observe the risk of recurrence and malignancy

4. Conclusion

Management of pelvic ABC was depending on the size of the lesion and stability in addition to clinical presentation. Early diagnosis and complete resection of tumor and treatment of pelvic ABCs should be planned based on location, size, and aggressiveness of the lesion, and relevant symptoms represent a viable treatment for this rare disorder to achieve increased life expectancy, low recurrence of the tumor and improved quality of life.

5. References

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