CASE REPORT

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Distal femur aneurysmal bone cyst treated with two stages of surgery in the pandemic COVID-19 by excision-skeletal traction and endoprosthesis replacement: a case report

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ABSTRACT

Introduction: An aneurysmal bone cyst (ABC) is benign but has a progressive nature that can cause severe problems if it occurs close to joints such as the knee. The ultimate goal of treatment is to remove the tumor, maintain limb function, and prevent a recurrence. Local therapy is still better than tumor resection. This study aims to describe a rare case about distal femur ABC treated with two stages of surgery in the pandemic COVID-19 by excision-skeletal traction and endoprosthesis replacement.

Case description: A 31-year-old woman presented with a lump on the right knee. The patient has complained of limping. Open biopsy was performed, and the patient was confirmed to have ABC on the distal of her right femur. The patient had a leg length discrepancy of 8 cm. We decided to do skeletal traction first. One week after the skeletal traction, the patient undergoes endoprosthesis replacement.

Conclusion: The management of wide excision of the tumor requires special postoperative attention. This is because aggressive measures can reduce the recurrence rate of tumors, but large reconstructions are also needed to maintain joint function.

Keywords: aneurysmal bone cyst, distal femur, surgery, endoprosthesis replacement
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INTRODUCTION

An aneurysmal bone cyst (ABC) was first discovered by Jaffe and Liechtenstein. This disorder is benign but has a progressive nature that can cause severe problems if it occurs close to joints such as the knee. Most ABC occurs in the bone metaphysis section.[1]

Research conducted by Kalim in 2019 at the Adam Malik Hospital in North Sumatra stated that the incidence of Aneurysmal Bone Cyst is in fourth place. While above it is inhabited by the three major primary bone tumors, namely sequentially; Osteosarcoma, Giant Cell Tumor, and Osteochondroma. These tumors attack more individuals in the third decade of age. Where the location is most often distal to the femur.[2]

ABCs are most often seen in children and young adults with no sex predilection. These lesions are lytic, usually eccentrically located and expansive with well-defined margins. There are blood-filled, separated by fibrous septa, with fibroblasts, osteoclast-type giant cells, and reactive woven bone.[1]

Management on ABC is already progressing. The ultimate goal of treatment is to remove the tumor, maintain limb function, and prevent a recurrence.[1,3] Local therapy is still better than tumor resection. Curettage was performed on ABC with the addition of an intralesional adjuvant to prevent a recurrence. Many ingredients as adjuvants such as; PMMA bone cement, argon beam, phenol, ethanol, and cryotherapy.[1,4]

However, tumor resection ("en bloc" surgical excision) promises an almost non-existent recurrence rate. By applying tumor resection, it is necessary to pay attention to the function of the associated joints. Where ABC often attacks bone metaphysis, a reconstruction plan (bone graft, arthrodesis, or joint replacement) is needed after resection. Other therapies such as; embolization, sclerotherapy, radiotherapy.[4]

Patients with ABC will complain of local pain, swelling, and an enlarged lump. Other complaints such as difficulty moving the joints and deformities can occur according to the severity of the tumor. If the ABC tumor reaches a level that is severe enough, it can fracture the bone. These tumors usually attack the femur, tibia, fibula, humerus, and axial bone in patients aged the second decade.[3]

The pathogenesis of these tumors is still being debated. Three concepts that can be accepted are; vascular, traumatic, or genetic. The concept of vascular explains

the presence of intramedullary primary vascular disorders that cause increased pressure and the formation of cysts in bone. While traumatic is a secondary vascular disorder due to previous trauma.[3]

Capanna et al. classified ABC into five morphologic subgroups, type I to type V ABC. Type I lesions are centrally located and well contained, with either no outline or a slightly expanded outline. Type II lesions have marked expansion and cortical thinning with the involvement of the entire bony segment. Type III lesions are eccentric and metaphyseal and typically involve only one cortex. Type IV lesions are the least common subgroup and develop subperiosteally, expanding away from the bone. Type V lesions occur periosteally and expand peripherally, ultimately penetrating cortical bone.[3]

As is well known, Aneurysmal Bone Cyst is a progressive benign bone tumor. This property should be noted because delaying therapy will provide sufficient time for ABC to grow and cause more bone destruction. In the end, the bones will become easier to fracture. Therapy, which can be done initially by injection of certain materials without mass resection, requires aggressive action, even with reconstruction. Therefore, this study aims to describe a rare case about distal femur ABC treated with two stages of surgery in the pandemic COVID-19 by excision-skeletal traction and endoprosthesis replacement.

CASE DESCRIPTION

We report a patient who came to the Surakarta orthopedic hospital. A 31-year-old woman presented with a lump on the right knee. The patient has felt this complaint since six years ago. The patient feels the lump as big as a chicken egg which is getting bigger.

Six years ago, the patient fell during activities at his home. After the incident, the patient complained of pain in the right knee. The patient underwent massage therapy. Complaints of pain disappeared, but the lumps began to appear. Patients can still stand and walk without using assistive devices.

For the last year, the patient has complained of limping. There is no history of trauma since the onset of the lump and limping. The patient went to a hospital in Cirebon. The patient underwent an MRI examination then the patient was referred to our hospital (Figure 1).

The patient underwent a follow-up examination then an open biopsy was performed on her right knee. Histopathological examination from the specimen showed multilocular spaces filled with numerous erythrocytes and few lymphocytes. The spaces were confined by woven bone fragments and connective tissue septae composed of fibroblast-like stromal cells lacking cytologic atypia.

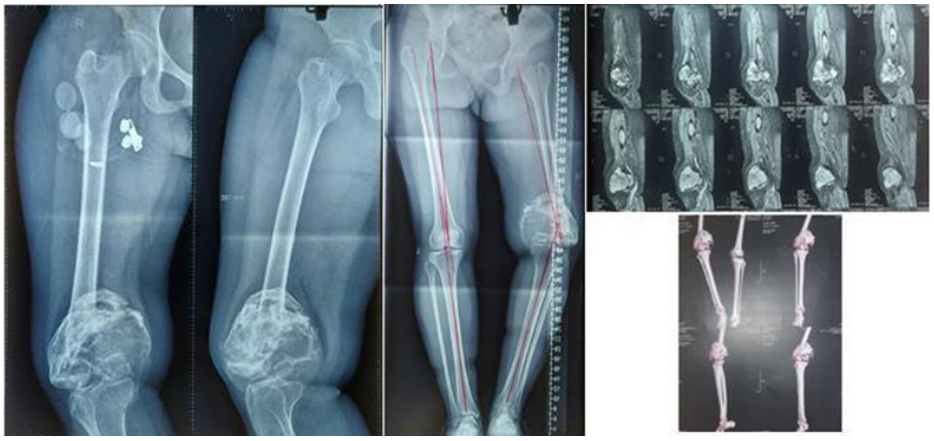

Figure 1. AP/Lat and full inferior extremity X-ray. MRI contrast and CT- scan 3D of the patient's right knee.

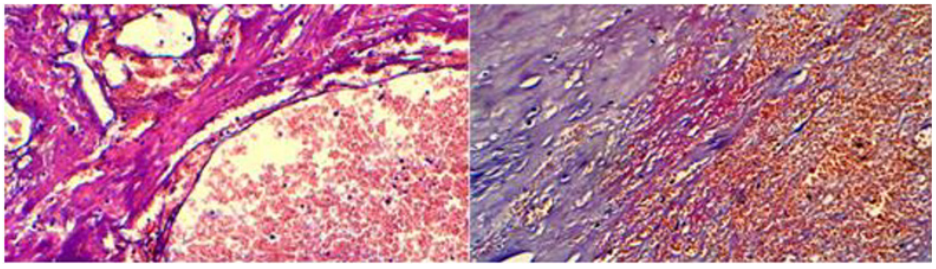

Figure 2. Histopathological examination from the specimen showed multilocular spaces filled with numerous erythrocytes and few lymphocytes.

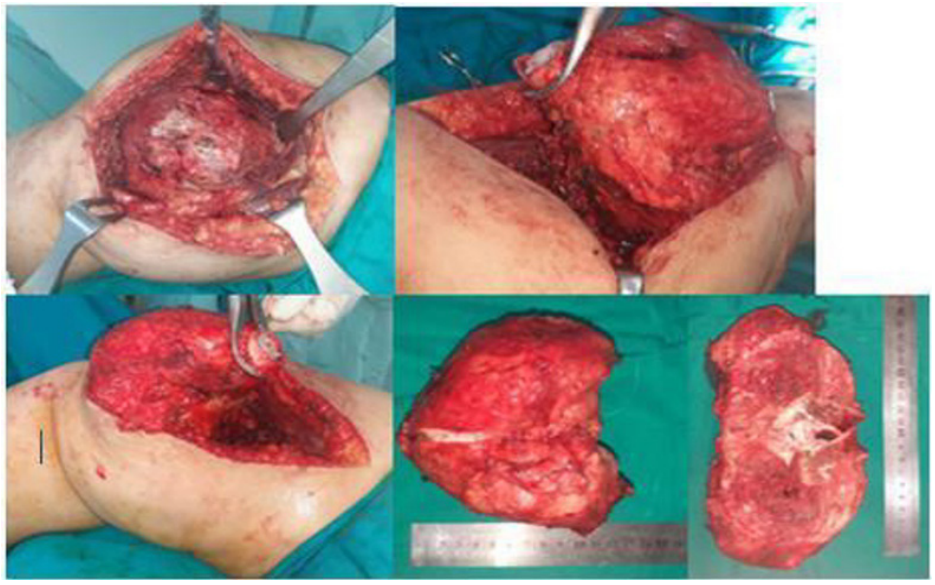

Figure 3. Excision of the tumor mass obtained a tumor size of 12x15 cm. When split in the middle, the size is 20cm.

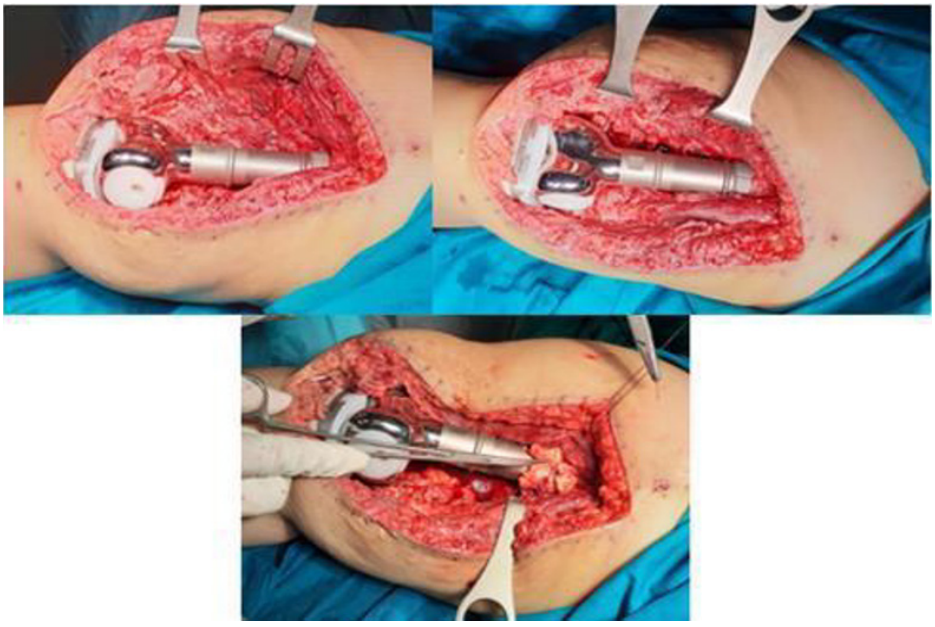

Figure 4. Megaprosthesis reconstructive surgery with applied bone grafts that were attached to the prosthesis and the bone junction.

Figure 5. Post-excision and post-mega prosthesis reconstruction procedure X-ray.

Figure 6. One-month postoperative follow-up. The patient can be mobilized using crutches.

Thus, histopathological features of the specimen supported the clinical and the radiological diagnosis of ABC (Figure 2).

The patient is planned for further surgery to remove the tumor. However, due to the global COVID-19 outbreak, patients failed to undergo the operation. One year later, the patient feels safe to return to treatment. Due to the long lag time of treatment, the patient had a leg length discrepancy of 8 cm. therefore, we decided to do skeletal traction first after tumor excision (Figure 3).

One week after the skeletal traction procedure, the patient's lower limbs were the same length. The patient was decided to undergo reconstructive endoprosthesis distal femur. Of course, with the condition of patients who have previously undergone tumor excision, the duration of endoprosthesis reconstructive surgery is shorter. We also applied bone grafts that were attached to the prosthesis and the bone junction. This action is expected to stimulate callus growth so that the connection between the prosthesis and bone will be stronger (Figure 4 and 5).

After three days of the second stage of the endoprosthesis, the patient began knee exercises. Physiotherapy is routinely performed on patients so that one-month postoperative follow-up can be mobilized with crutches (see Figure 6).

DISCUSSION

In our case, ABC occurred in a middle-aged woman located in the distal femur. This is in line with the statement that it affects between the ages of 20 and 40 years. ABC also affects the long bone around the knee more often.[3,5] The patient began to complain of pain in the right knee after the fall. This mechanism is related to the traumatic pathogenesis of ABC. The pain felt by the patient was continuous and was not affected by movement. This complaint

comes from bone destruction. The soft tissue becomes swollen and edema is a sign of progression of bone destruction from outside the bone. Because ABC occurs near the joint, it will interfere with joint motion, joint effusion, and synovitis.[3]

However, patients can still mobilize without assistive devices until the last year the patient had difficulty mobilizing. Six years is sufficient time for the tumor to develop and become osteolytic to result in a fracture.[6] No sources have stated the length of time it would take ABC to cause a pathological fracture. This is closely related to the osteolytic process by ABC.[6] Differential diagnoses for ABC include the brown tumor of hyperparathyroidism, giant cell tumor (GCT), telangiectatic osteosarcoma, and malignant fibrous histiocytoma, fibrous metaphyseal defects, giant-cell-rich osteosarcoma, metastatic cancer, chondroblastoma, chondromyxoid fibroma, and non-ossifying fibroma.[6]

Management in this patient is "en bloc" resection followed by endoprosthesis replacement of the distal femur. ABC treatment is the percutaneous injection of substances such as PMMA bone cement, argon beam, phenol, ethanol, and cryotherapy.[4] Osteosynthesis is carried out only when needed.[6]

Due to the time lag of one year, the LLD was 8 cm. Patients postponed surgery because they waited for the pandemic to subside. We decided to do skeletal traction first, then continue the second stage of endoprosthesis replacement.[5] During the endoprosthesis insertion, we performed extracortical bone fixation (bone graft affixed to the porous segment). This aims to create a callus between the bone and the implant to be more fixed in place.[5]

Croci et al. Studied 37 cases of primary bone tumors such as aneurysmal bone cyst (ABC) under reconstructive endoprosthesis. They got 56.8% of cases with good results. In this study, evaluation was carried out based on several criteria such as; the presence of pain, range of motion, reconstruction stability, surgical and oncologic complications, and patient acceptance.[7]

Another study was conducted by Rubio et al. On 22 patients who underwent tumor reconstructive endoprosthesis. The study found the problem of infection was still a major cause of revision surgery. Some of the other problems that followed such as; aseptic loosening, structural failures, soft tissue failures, and tumor progression.[8]

A case report by Ulici et al on a case of an aneurysmal bone cyst (ABC) in an 11-year-old child. After embolization and biopsy, an aneurysmal bone cyst was found in the proximal femur. Thirty weeks is sufficient time for these tumors to develop aggressively. The team of doctors decided to do tumor resection and reconstructive endoprosthesis. The evaluation results in these patients found good results with complete recovery of function and no local relapse.[9]

CONCLUSION

The management of wide excision of the tumor requires special postoperative attention. This is because aggressive measures can reduce the recurrence rate of tumors, but large reconstructions are also needed to maintain joint function. As long as there is no need for prosthetic replacement or osteosynthesis, percutaneous injection is a good therapeutic option for ABC because it avoids impaired joint function.

DISCLOSURES

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In this case report, we use our resources without any financial assistance from other parties.

Conflict of Interest

There is no conflict of interest from authors in this case report publication.

Author Contribution

First author involved in conceiving, designing, and supervising the manuscript. Second conduct the study analyze the data. All authors prepare the manuscript and agree for this final version of the manuscript to be submitted to this journal.

Ethical Statement

In this case report, we have obtained permission from the patient to conduct scientific publications in the form of photos and others. Information regarding the patient's identity is not explained in this paper.

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