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Case Study

Management Aneurysmal Bone Cyst Using Methylprednisolone Sclerotherapy: A Case Series

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Abstract: The aneurysmal bone cyst (ABC) is a benign intraosseous or rare tumour-like lesion. The ABC therapy methods have evolved over time. However, the best management is still debatable. Surgical curettage shows high recurrence rates ranging from 30% to 50%. In the recent decade, Percutaneous sclerotherapy has developed as an excellent therapy technique for aneurysmal bone cysts. Therefore, this study aims to report the clinically and radiologically outcome using methylprednisolone in patients with ABC. There are two cases in this study. A male, 26 years old, complained about pain in his left hip one year ago. From a biopsy, we found an immunophenotype that is relevant to aneurysmal bone cysts with cystic spaces and stromal giant cells. Therefore, we diagnosed the patient with Aneurysm Bone Cyst Trochanter Left Femur. Secondly, a patient 22-year-old male has complained of pain in the left foot for six months. We got only red blood cells with lymphocytic inflammatory cells and without malignant cells from an open biopsy. We diagnosed patients with an aneurysmal bone cyst on the left calcaneus bone. All patients were treated with methylprednisolone on their lesion twice. All of the tumours subsided during follow-up and significant pain reduction.
Keywords: Aneurysmal bone cyst, sclerotherapy, methylprednisolone.

Introduction

The aneurysmal bone cyst (ABC) is a benign intraosseous or rare tumor-like lesion that was initially identified in 1942 by Jaffe and Lichtenstein.[1] The aneurysmal bone cyst (ABC) typically manifests during the first decades of life, with a prevalence of 1.4 per 100,000, contributing to around 1% of benign bone tumors.[2]

ABC commonly occur in the metaphysis of the long bones and column vertebrae, and ABC has characteristic locally aggressive tumors with a high capacity for local reccurence.[3,4] ABCs' pathophysiology remains unclear, with hypotheses varying from a post-traumatic reaction, vascular malformation, and, recently, gene abnormality.[5]

Patient ABC commonly complains of swelling, pain, and deformity in the tumor area. Pathological fracture is also occasionally apparent. A well-defined osteolytic, "expansile" lesion with likely periosteum blowout and a soap bubble appearance can be seen on plain radiography.[6,7] However,

most studies considered biopsy indispensable, and up to this point, it has been the gold standard assessment.[8] Before deciding on treatment, histopathological confirmation of ABC is indispensable to differentiate from telangiectatic osteosarcoma, which may have some resemblance radiologically.[9,10]

The ABC therapy methods have evolved over time; however, the best management is still debatable. There are several options from curettage (with or without polymethylmethacrylate filling of the cavity) or autologous en block excisions with the repair of the bone defect with bone substitution material, selective embolization of the feeding arteries, radiation therapy, and cryosurgery.[8,11]

Surgery by curettage is the standard care in ABC patients.[12] Unfortunately, in most cases, resection is not a possibility. Thus, intralesional surgeries[9] and surgical curettage show high recurrence rates ranging from 30% to 50%.[9,13] Therefore, alternative management such as prolotherapy is needed to explore optimal management and reduce recurrence in ABC patient.[4,14]

In the recent decade, Percutaneous sclerotherapy has developed as an excellent therapy technique for aneurysmal bone cysts.[14] Previously, various sclerotherapy materials have been reported, such as polidocanol,[14] Ethibloc,[3,15,16] Calcitonin, and methylprednisolone,[17] Alcohol.[18] However, till now, few studies reported methylprednisolone only for patients with ABC. Therefore, this study aims to report the clinically and radiologically outcome using methylprednisolone in patients with ABC. This article has followed CARE checklist and guidelines.

Presentation of Case

Case 1

A male, 26 years old, came to the orthopedic clinic to complain about pain in his left hip for one year and getting worse in the last three months. Pain worsens when a patient is doing strenuous activity. The pain was localized and did not radiate. The patient is currently able to walk. The patient has been an active smoker for 13 years ago.

Patients had experienced trauma after traffic accidents, but no fracture occurred about five years ago. The patient routinely takes painkillers two times a day but still feels pain. From the physical examination, we found tenderness (+), and pain (+). On the other hand, the range of motion was normal and distal neurovascular and within normal limits.

Then, we did a radiological examination of the left femur and found osteolytic lesions of the left femur neck-trochanter major-intertrochanteric bone femur suggestive of benign aneurysmal bone cysts (Figure 1A).

To establish a diagnosis, we performed a histological examination using an open biopsy. We obtained an immunophenotype from a biopsy that is relevant to aneurysmal bone cyst with cystic spaces and stromal giant cell (Figure 2). Therefore, from all of the examinations carried out, we diagnosed the patient with Aneurysmal Bone Cyst Trochanter Left Femur. Then, the patient was treated with an injection of Depo-Medrol® (methylprednisolone) in tumor lesions three times with a 1-month interval for each injection.

After the last injection, we evaluated clinical and radiological outcomes for two weeks after injection (Figure 1B). The patients expressed that the pain significantly reduced from VAS score from 5 to 1, and from radiological examination, there was a reduction in the size of the tumor lesions after injection using Depo-Medrol® 40 mg. There were no side effects that occurred during the injection of Depo-Medrol®.

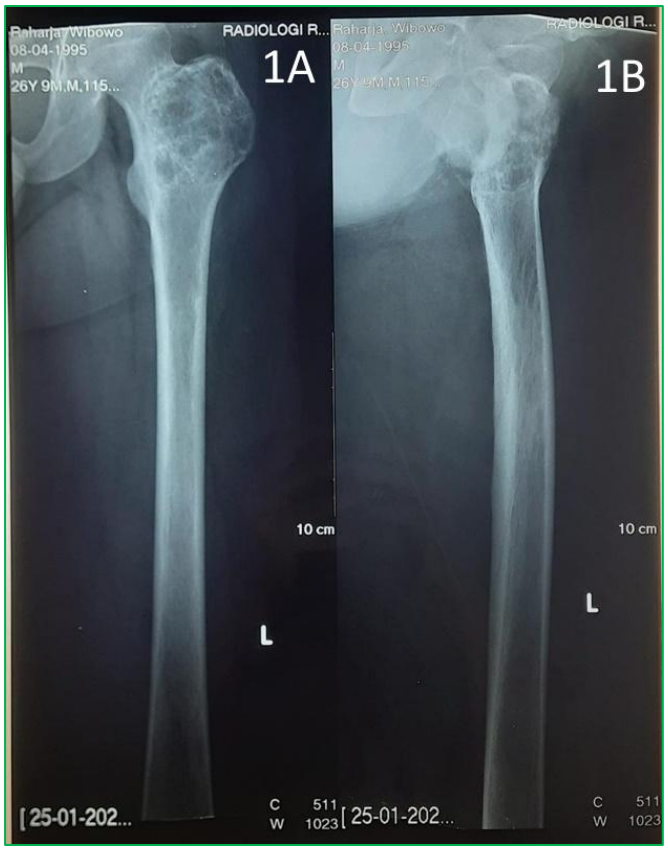

Figure 1. X-ray on left femur before and after injection

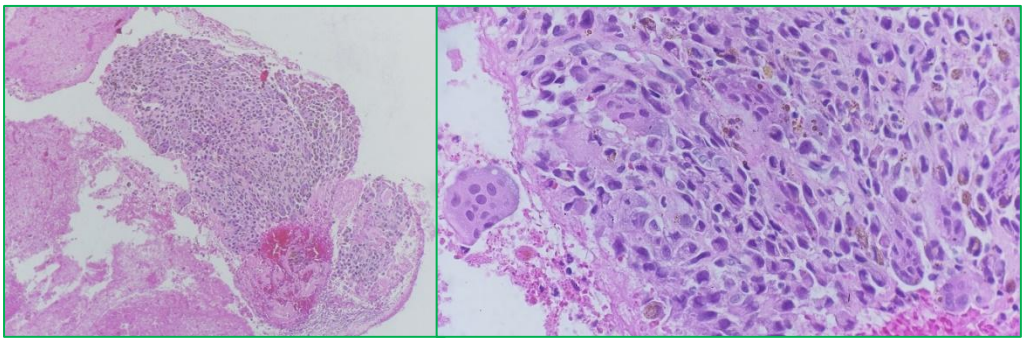

Figure 2. Cystic spaces and stromal giant cells

Case 2

Twenty-two-year-old male came to an outpatient clinic with chief complaints of pain in the left foot for six months ago after patient does jogging. Initially, the type of pain is intermittent pain. However, his pain has been getting worse and permanently in the last week especially when patients is walking.

The patient has a history of trauma after a motorcycle accident with the left foot being hit by a motorbike five years ago. There was no history of surgery, and there was no pain or mass in another area. The patient's routinely taking painkillers once a day, but the symptom doesn't reduce. From Physical examination, there was tenderness (+), pain (+), but limited range of movement was normal (Active Ankle plantar-dorso flexion: 30°-50). Then, we performed a Plain x-ray left calcaneal ap/lateral and obtained lytic lesion of the calcaneus suggestive benign (Figure 3A). We also performed MRI ankle and we found primary bone tumor, cystic with an internal hemorrhagic component of the suspected left calcaneus ABC. For confirmation diagnoses, we also underwent histological examination using open biopsy. From an open biopsy, we got only red blood cells with lymphocytic inflammatory cells and without malignant cells found. We diagnosed the patient with an aneurysmal bone cyst on left calcaneus bone. Then, we treated patients with Depo-Medrol® 40mg

(methylprednisolone) in the area of the tumor twice with an interval of one month. Furthermore, an evaluation was carried out one week after 1-week post the second injection and obtained satisfactory results (Figure 3B). The patient stated that the pain was reduced, and the results of the x-ray evaluation showed that the tumor lesion on the left foot had reduced. There is no adverse effect that has been reported.

Figure 3. X-ray on left foot before and after injection

Discussion

Since its discovery in 1942, the pathophysiology of ABCs is still debatable. Historically, most researchers assume it is a natural reaction to a specific change in hemodynamics that causes a significant increase in venous pressure and the formation of a dilated capillary network within the affected bone.[19] Until 1999, Panoutsakopoulos et al[20] reported the clonal theory that there is an unbalanced chromosomal translocation t(16;17)(q22;p13) as a cytogenetic anomaly in primary aneurysmal bone cyst.

An aneurysmal bone cyst is a tumour that has sparked disagreement, particularly beyond its classification and treatment.[12–14] ABC is a benign but locally aggressive lesion in which there are no clear primary treatment options. Due to the obvious consequent disability and the needed surgically reconstruct in the context of benign disease, En Bloc resection is not an option in this case.[12]

Management of ABC using Intralesional curettage with grafting or without grafting is still the most widely used procedure.[14,21] However, this technique reported recurrence rates are high, between 10% to 59%. Moreover, surgical procedures often cause problems such as the risk of massive bleeding and also anatomic instability.[22] Therefore, in the last decade, the concept of curettage management is gradually shifting to sclerotherapy treatment. Several managements have been reported that sclerotherapy treatment promises a good outcome. However, the optimum agents are currently being debated.

There are various types of sclerotherapy reported to be used in percutaneous injections to treat ABCs. Several studies have stated that polidocanol and ethibloc are the optimum choices for sclerotherapy due to minimizing side effects with a low recurrence rate.[3,14-16] However, not all hospitals, especially in Indonesia, have these drugs. Therefore, methylprednisolone can be used as an alternative for patients with ABC.[17] Other drug options that have been reported to be used include calcitonin, methylprednisolone, alcoholic zein, bisphosphonate, bone marrow aspirate, and doxycycline. Even though the evidence is either restricted to case reports or small studies, the outcomes are inconsistent.[7]

In these two case studies, both results were quite satisfactory during the follow-up process. This finding related to three previous studies that have reported using methylprednisolone to treat aneurysmal bone cysts. The difference between this study and previous studies is that they used methylprednisolone and calcitonin in the previous study, whereas, in this study, only methylprednisolone was used.[23–25] Methylprednisolone is mostly used for a simple bone cyst. In contrast, many previous studies reported that treatment with steroid injections offers acceptable results in simple bone cysts. Several studies reported that steroid has an angiostatic effect that inhibits osteoclast activity.[17] Therefore, the potential that exists in the use of steroids on ABC patients can be used for the case of this patient.

Conclusion

This study shows that non-invasive methylprednisolone administration is as effective in aggressive ABCs, has no adverse effects, and has a potential rapid response with minimally invasive treatment. As a note, Therapy should begin after a complete radiological examination and histological confirmation of the diagnosis as soon as possible to achieve a good outcome. This study has several limitations. Firstly, a short follow-up period is still lacking to be able to conclude the results properly. A more significant number of patients and longer follow-up can be considered for further research. The results of this investigation show that the use of methylprednisolone in patients with ABC gives satisfactory results, with rapid results and promising outcomes.

Conflicts of interest: The authors declare no conflicts of interest.

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