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Case Report
Use of Triamcinolone Acetonide as Sclerosing Agent in the Treatment of Aneurysmal Bone Cyst: A Case Report

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Abstract

Background and Objective: Aneurysmal bone cyst (ABC) is classified as a benign and non-neoplastic bone lesion, which can be filled with multiple cavities containing blood. It is a locally destructive lesion. Manifestation is more than 50% in long bones and mostly seen before the age of 20 years in 80% cases. As an expansive and osteolytic lesion, even though benign, it may lead to pathological fracture. This study aimed to investigate the use of Triamcinolone acetonide as sclerosing agent. **Materials and Methods:** This case report described a 20 year-old female with a diagnosis of aneurysmal bone cyst of right proximal femur. Minimally invasive treatment was performed with percutaneous injections of triamcinolone acetonide as sclerosing agent. **Results:** At three years follow up, patient had resumed to high impact exercise and the radiograph showed a fully resolved lesion. **Conclusion:** Triamcinolone acetonide may be used as a single sclerosing agent in developing country where more known agents are difficult or too expensive to obtain.

Key words: Aneurysmal bone cyst, triamcinolone acetonide, manifestation, percutaneous injection, sclerosant agent

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Competing Interest: The authors have declared that no competing interest exists.

Data Availability: All relevant data are within the paper and its supporting information files.

INTRODUCTION

Aneurysmal bone cyst (ABC) is described as an expansile, osteolytic, locally destructive and rapid growing tumor-like lesion[1]. It may be filled with blood and had walls, separating each "bubble", which may contain giant cells with osteoid tissue and osteoclast-like characteristics. About 20% of cases arise in appendicular skeleton and may be identified as a primary lesion or secondary lesion. Pain as a major chief complaint led patients with ABC to seek treatment[1-4].

The treatment of ABC had been done by several methods. The surgical treatment of ABCs typically involves curettage with or without bone grafting[5,6]. However, there had been increasing trend of minimally invasive procedures to avoid complications of open surgical procedures[7,8].

Minimally invasive procedure including in tralesional injection of sclerosing agent had gained attention in the treatment of ABC. Sclerosant works by causing swelling on blood vessel and cutting off the flow of blood, resulting in shrinkage of the vessels with the help of image guiding or USG guiding for an accurate delivery of sclerotherapy[4,7]. Some sclerosants used for the treatment of ABC found in literatures are ethibloc, denosumab, polidocanol, calcium sulfate, absolute alcohol and combination of calcitonin and methylprednisolone[1,9-13]. Almost all mentioned sclerosing agents are costly and not readily available in developing country. The aim of this study was to examine the use of triamcinolone acetonides sclerosing agent in the treatment of aneurysmal bone cyst.

In this study, the use of triamcinolone acetonide was suggested as sclerosing agent in the treatment of ABC. This case report could provide additional information regarding the choices of sclerosing agent that can be used for the treatment of ABC.

MATERIALS AND METHODS

The case report was done in Orthopaedic Department at Wahidin Sudirohusodo Hospital in Makassar, Indonesia with a time duration of 37 months, starting from April 2015 to May 2018.

Patients and methods: Parameter of this study was functional outcome of the subject of this study. Observations were done through follow ups.

A 20-year-old female presented with right hip pain for several months after she slipped and fell on her right buttock. She felt increasing intermittent pain on her right hip for as long as 5 months with consumption of pain killer. She was a climber and a high-impact sport woman before the accident and she was not able to resume to her normal activity due to her pain. She stated that the pain was intermittent, with no worsening at night. She also denied weight loss or fever. There was no history of respiratory tract infection and was not in consumption of any drug or medication. There was no history of trauma after the incident.

On physical examination, there was marked swelling and tenderness on her right hip, but range of motion was within normal limit. No sign of inflammation and deformity was present and neurovascular assessment was within normal limit. On gait examination, patient was limping when her right lower extremity was on weight bearing. Palpation of groin region revealed no enlargement of lymph nodes.

Radiographic examination revealed an expansile lytic lesion on the right trochanter area, with thin rim of periosteal new bone, a narrow zone of transition, no matrix mineralization and no soft tissue involvement (Fig. 1, X-ray of pelvic), her blood examination were within normal limits of hospital. Erythrocyte sedimentation rate was not elevated and C-reactive protein was non-reactive. Lactate dehydrogenase, alkaline phosphatase and total serum calcium were also within normal result.

Biopsy was performed and pathology anatomy smears were predominated by large amount of erythrocytes with scattered macrophages. No malignant cells were found (Fig. 2, PA of lesion).

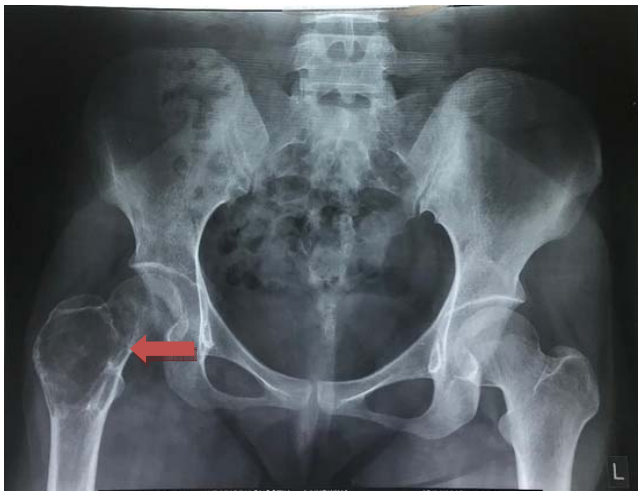

Fig. 1: Anteroposterior view of pelvic radiograph showed eccentric multiple lesions on right intertrochanter region, with geographical type of destruction, no zone of calcification, periosteal reaction, matrix and soft tissue involvement

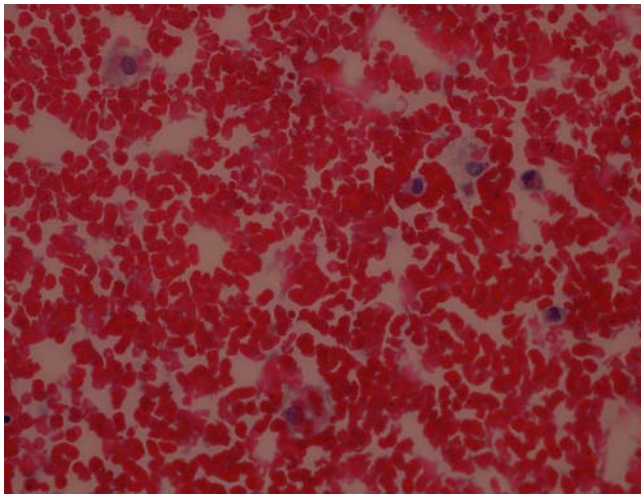

Fig. 2: Smears of samples taken from the lesion, inspected from pathology anatomy. Predominated by large amount of erythrocytes and scattered macrophages were observed. No malignant cell was found

A clinical pathological conference was held to discuss the case with end result of aneurysmal bone cyst. Intralesional injection was chosen as method of treatment, using a single dose triamcinolone acetonide 40 mg. An 11 gauge aspiration needle was inserted to aspirate the content of the cyst and when aspiration was finished 40 mg of triamcinolone acetonide was injected into the cyst. The procedure was one time and patient was discharged afterwards.

RESULTS

Subject of this case report came with pain in right lower extremity and upon radiograph examination, a lesion was revealed as seen in Fig. 1. Further examination was performed with a fine needle biopsy as chosen methods. Results of biopsy was in line with aneurysmal bone cyst (Fig. 2). At 3 months of follow up, patient reported her return to normal daily activity routine without any pain. However she was still unable to perform high impact training. As shown on Fig. 3, sclerosis began to appear on top of the lytic lesion, slowly replacing it. After a period of 3 years of follow up, it was found from the single injection of triamcinolone acetonide that the patient was able to return to normal daily activity with high impact sport without residual pain reported (based on auto-anamneses) and on X-ray findings, the lesion had fully disappeared and sclerotic area (as seen in black circled area in Fig. 4) had appeared, replacing the lytic lesion.

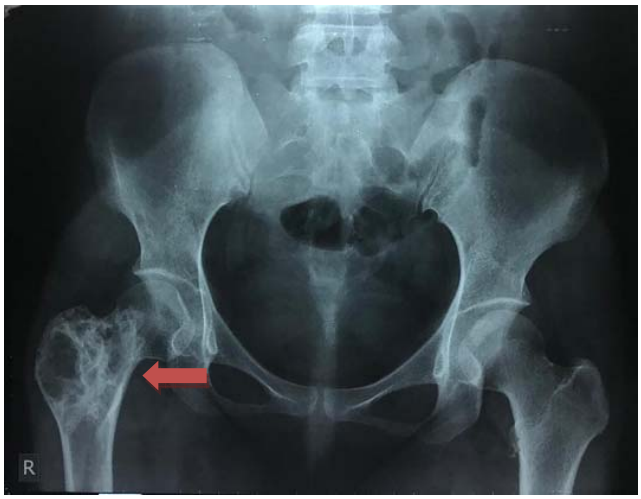

Fig. 3: Anteroposterior view of pelvic radiograph at 3 months follow up which revealed increasing sclerosis within the cysts on right intertrochanter region

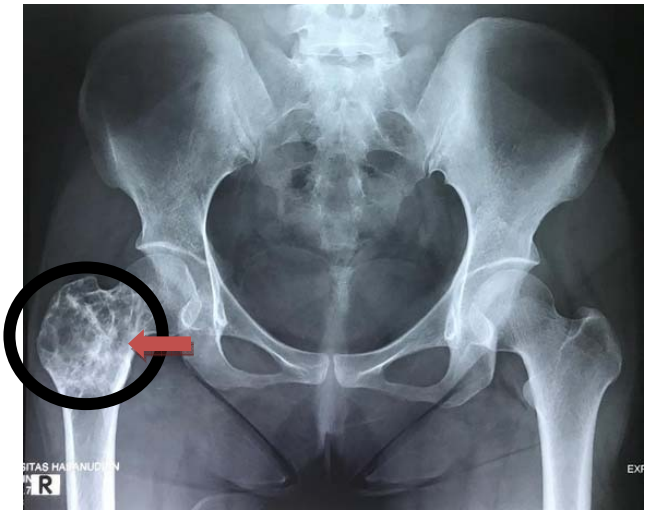

Fig. 4: Anteroposterior view of pelvic radiograph taken at 3 years follow up, showed fully resolved lesion and increased sclerotic on right intertrochanter region

DISCUSSION

Aneurysmal bone cysts incidence rate is highest on childhood and early adulthood (up to 80% cases are seen below the age of 20 years)[1,4]. The incidence for male and female had a slight predominance to female with mean age of incidence at 19.7 years. Taking into account the patient in this case was a 20 years old female with a pain history of 5 months[1,12].

Plain radiograph may reveal expansile osteolytic lesion (Fig. 1). Biopsy with the chosen method being Fine Needle Aspiration should be performed and sufficient quantity of cells from the site of the lesion is taken and analyzed[8]. Blood filled spaces and septa, not lined by endothelial cells with giant cells were usually found in the stroma of the tumor, much like the one found in patient[4,12].

Due to the osteolytic and expansile characteristic of the lesion, treatment is preferred over mere observation. Treatment ranging from non-invasive to invasive consists of en bloc surgical excision, intralesional surgical procedure with or without local adjuvants, minimal invasive surgical techniques, embolization, sclerotherapy and injection of drugs and radiotherapy[4-6]. The current trend of treatment had been heavily focused on sclerotherapy, with less and less amount of injection performed and variation of sclerotherapy agent[4].

Injection of drugs intralesionally is preferred because it offer a least invasive therapeutic option. It works by causing damage to vascular endothelium causing cascade chain of events resulting in healing of the lesion. Some of the more common agents injected is calcitonin, which is known to inhibit osteoclastic activity and promotes trabecular bone formation and methylprednisolone, which was believed to have anti-angiogenesis effect[1,4,7,9].

In this study, only triamcinolone acetonide was use, which is a stronger type of corticosteroid, compared to methyl prednisolone, which enabled complete resolution of pain and lesion in the affected area. Surprisingly incurrent findings, complete remission of the disease within 3 years was found, with only a single injection of a more potent type of corticosteroid. With the chosen sclerosing agent and time of remission, current results were comparable to those performed by Oliveira *et al*, in which the time of first healing and complete remission was comparable in 2.8 and 37.7 months[1]. This study was also comparable to the results of other study on calcitonin and steroid in which the amount of injections required was similar, although, follow up time was shorter for the agent combination[2].

Current sclerotherapy agent showed better results without residual pain when compared to a case report by Rai and Collins[7] even though complete remission was achieved faster at 6 months after treatment by this study. A Chinese study using absolute alcohol was performed in 2015. And even though after 2 years follow up partial healing occur and improvement was noted, multiple injection was needed and great precaution was needed as stated by the author[10].

Denosumab was also used in a study and provided volume reduction and decreased clinical symptoms. However, the amount of patient visitation and multiple doses was needed with some side effects, including hypercalcemia, suppressed parathyroid hormone and even relapse of the disease. The chosen sclerotherapic agent was considered far more superior, even though more research is needed[11,12]. Sclerotherapy using surgiflo and alcohol by Ghanem *et al*.[3] showed good result, however, the subjects required up to 71 months of follow ups with varying clinical results. Polidocanol injection gave good results following intralesional injection with a comparable follow up time, however multiple visits was required for most of the subjects of the study[13].

Another comparable result comes from a research study which combined ethibloc, aetoxisclerol and liquid absolute alcohol. However the use of absolute alcohol created skin necrosis due to leakage and ethibloc in their case created inflammatory reaction[14]. Comparing this to subject of this report whom had shown very little to no adverse effect, in author's opinion, warrants further research in the hope of creating a new shift of sclerosing agent of choice. Current result is comparable to those performed by George *et al*.[9] in which consolidation of bone and re-constitution of bone is achieved, but without the side-effect of leakage of contrast.

A research by Zarzour *et al*.[15] used CT guided radio frequency ablation, but some of subjects needed cementation, resulting in more procedure performed. After 1 year all subjects of the study shows complete remission and no pain was reported. The use of MSCT as guide was meant to help in detecting the exact location of the lesion, however, resulting in non-outpatient procedure.

A more thorough study would be needed to establish a good evidence-based footing for single injection of drugs, instead of the usual repetitive visits with multiple drug injection.

CONCLUSION

While diagnosis of ABC may follow a pre-determined course, treatment may vary from observation, intralesional injection to curettage of lesion, with a shift of trend on the chosen treatment from the more invasive to the less or minimally invasive treatment. This case report presented a successful treatment with good functional outcome using only a single dose of triamcinolone acetonide.

SIGNIFICANCE STATEMENTS

Minimally invasive procedure including intralesional injection of sclerosing agent have gained attention in the treatment of ABC. Some sclerosants used for the treatment of ABC that we found in literatures are ethibloc, polidocanol and

combination of methylprednisolone-calcitonin. In our case, it suggested the use of triamcinolone acetonide as sclerosing agent in the treatment of ABC. To our knowledge, the use of triamcinolone acetonide as sclerosant in the treatment of ABC has not yet been published in any literatures. In our opinion, this case report could provide additional information regarding the choices of sclerosing agent that can be used for the treatment of ABC.

REFERENCES

1. Oliveira, M.B.D.R., W. Meohas, G.S. de Carvalho, R.R. Rodrigues and F.C.R. Oliveira *et al*., 2017. Primary aneurysmal bone cyst of long bones treated with a single dose of calcitonin and methylprednisolone percutaneous intralesional injection: A case series and literature review. J. Orthop. Oncol., Vol. 2. 10.4172/2472-016X.1000113.
2. Chang, C.Y., S.V. Kattapuram, A.J. Huang, F.J. Simeone, M. Torriani and M.A. Bredella, 2017. Treatment of aneurysmal bone cysts by percutaneous CT-guided injection of calcitonin and steroid. Skeletal Radiol., 46: 35-40.
3. Ghanem, I., N. Nicolas, M. Rizkallah and S. Slaba, 2017. Sclerotherapy using surgiflo and alcohol: A new alternative for the treatment of aneurysmal bone cysts. J. Child. Orthop., 11: 448-454.
4. Tsagozis, P. and O. Brosjo, 2015. Current strategies for the treatment of aneurysmal bone cysts. Orthop. Rev., 7: 106-110.
5. Krishna, L.G., K. Pandey and A. Agarwalla, 2018. Aneurysmal bone cyst of patella treated with curettage and cementing-A case report. Int. J. Scient. Res., 7: 47-48.
6. Reddy, K.I.A., F. Sinnaeve, C.L. Gaston, R.J. Grimer and S.R. Carter, 2014. Aneurysmal bone cysts: Do simple treatments work? Clin. Orthop. Related Res., 472: 1901-1910.
7. Rai, A.T. and J.J. Collins, 2005. Percutaneous treatment of pediatric aneurysmal bone cyst at C1: A minimally invasive alternative: A case report. Am. J. Neuroradiol., 26: 30-33.
8. Peabody T.D. and S. Attar, 2014. Orthopaedic Oncology: Primary and Metastatic Tumors of the Skeletal System. Springer International Publishing, Switzerland.
9. George, H.L., P.N. Unnikrishnan, N.K. Garg, J.S. Sampath, A. Bass and C.E. Bruce, 2009. Long-term follow-up of Ethibloc injection in aneurysmal bone cysts. J. Pediatr. Orthop. B, 18: 375-380.
10. Han, Y.F., X.D. Fan and L.X. Su, 2015. Percutaneous sclerotherapy with absolute alcohol to treat aneurysmal bone cyst of the frontal bone. J. Craniofac. Surg., 26: 456-458.
11. Kurucu, N., C. Akyuz, F.B. Ergen, B. Yalcin and K. Kosemehmetoglu *et al*., 2018. Denosumab treatment in aneurysmal bone cyst: Evaluation of nine cases. Pediatr. Blood Cancer, Vol. 65, No. 4. 10.1002/pbc.26926.
12. Palmerini, E., P. Ruggieri, A. Angelini, S. Boriani and D. Campanacci *et al*., 2018. Denosumab in patients with aneurysmal bone cysts: A case series with preliminary results. Tumori J. 10.1177/03008916118784809.
13. Brosjo, O., P. Pechon, A. Hesla, P. Tsagozis and H. Bauer, 2013. 2013. Sclerotherapy with polidocanol for treatment of aneurysmal bone cysts: Good results in 37 of 38 consecutive patients. Acta Orthop., 84: 502-505.
14. Batisse, F., A. Schmitt, T. Vendeuvre, D. Herbreteau and C. Bonnard, 2016. Aneurysmal bone cyst: A 19-case series managed by percutaneous sclerotherapy. Orthop. Traumatol.: Surg. Res., 102: 213-216.
15. Zarzour, O.A., F.R. Santiago, N.O. Serrano, A.H. Abdallah, M.A. El-Sharkawy and A.F. Mourad, 2018. CT-guided radiofrequency ablation in patients with aneurysmal bone cysts. Eur. J. Radiol., 100: 116-123.