Aneurysmal bone cyst in the proximal femur: Combined approach of management: A case report

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Abstract

Purpose of Study: To study the effectiveness of combined extended curettage and sclerosant therapy in the management of aneurysmal bone cyst of proximal femur.

Materials and Methods: A 22 years old male presented with complaints of left hip pain since 20 days, was examined clinically and radiologically. On MRI, he was diagnosed to have a non-expansile lytic lesion in the left proximal femur with features suggestive of aneurysmal bone cyst or an unicameral bone cyst. Patient was planned and taken up for aspiration and curettage followed by a single sitting sclerosant therapy with 3% polidocanol, bone cementing combined with plating. Curetted sample was sent for histopathological examination. Patient was advised restricted weight bearing for a period of 4 weeks. Regular follow-ups of the patient was done at 1, 3, 12 and 18 months post-operatively to assess for recurrence.

Results: Histo-pathological examination confirmed the diagnosis of aneurysmal bone cyst. With regular follow-ups, patient had a good functional outcome and no evidence of recurrence with this technique at 18 months follow-up.

Conclusion: Aneurysmal bone cyst has a higher recurrence rate with curettage and bone grafting alone. Extended curettage along with sclerotherapy with 3% polidocanol has shown no evidence of recurrence at 18 months follow-up in our study. Hence, this combined approach can be recommended in the management of aneurysmal bone cyst.

Key words: giant cell tumor, unicameral bone cyst, femur, sclerosant, curettage

Introduction

Aneurysmal bone cysts (ABCs) are benign expansile lesions produce cavities within the bone that fill with blood and are lined by proliferative fibroblasts, giant-cells, and trabecular bone, arising predominantly in the pediatric population that can cause local pain, swelling, and pathologic fracture [1, 2]. ABCs are commonly seen during childhood and young adulthood with a median age of 13 years, and 90% of lesions are found prior to age 30. Females are slightly more affected with an estimated M:F sex ratio of 1:1.16[3]. ABCs also occur secondary to other bone tumors such as chondroblastomas, giant cell tumor, chondromyxoid fibroma, non-ossifying fibromas, or fibrous dysplasia. These secondary ABCs account for nearly 30% of all ABCs, and they are not considered a neoplasm because there is no known translocation or genetic aberrancy is present [4]. ABCs have a predilection for the metaphysis of long bones including the femur, tibia/fibula, and upper extremity. Even though X-rays and MRI are used for diagnosis, histo-pathological examination of biopsy specimen still remains as gold standard.

The standard of treatment remains curettage and grafting to fill the bone void, but the adjuvant or alternative treatment methods to reduce recurrence are numerous including phenol cauterization, cryosurgery, argon beam coagulation, etc. Embolization of the feeding arteries has been suggested as an alternative with good outcomes, but the technique is technically demanding as some lesions may lack a large afferent vessel [5]. Recent data suggest that percutaneous sclerotherapy is a safe alternative to surgery for treatment of ABCs [6]. But, most often the patient treated with sclerotherapy alone, end up with recurrence and go into surgical resection and bone grafting. Hence, only in surgically inaccessible regions the use of sclerotherapy has gained popularity.
Aim of the Study
To study the effectiveness of combined extended curettage and sclerosant therapy in the management of aneurysmal bone cyst of proximal femur.

Case Report
A 22 years old male presented with complaints of left hip pain since 20 days, which was insidious in onset, dull-aching, non-radiating type of pain, present throughout the day. Pain aggravated on strenuous activity. The patient approached the orthopaedician as the pain started impending his daily routine. He was examined clinically and radiologically; and was diagnosed to have a non-expansile lytic lesion in the left proximal femur with features suggestive of aneurysmal bone cyst or an unicameral bone cyst from an MRI study. Patient was planned and taken up for aspiration and curettage under C-arm guidance along with a single sitting sclerosant therapy with 3% polidocanol, followed by bone cementing combined with plating. Curetted sample was sent for histo-pathological examination. Histo-pathological examination showed multinucleated giant cells with reactive woven bone and confirmed the diagnosis of aneurysmal bone cyst. Patient had no intra-operative or post-operative complications. Patient was advised restricted weight bearing for a period of 4 weeks post-operatively, after which complete weight bearing and mobilization was started. Regular follow-ups of the patient were done at 1, 3, 12 and 18 months post-operatively to assess for recurrence.
Discussion
Aneurysmal bone cysts located in the peri-articular locations of long bones pose a risk of iatrogenic fractures during aggressive curettage. Despite best efforts at curettage, studies have shown highly variable recurrence rates of up to 59% [7]. However, recurrence rates have significantly reduced with the use of effective local adjuvants like bone cement, use of high-speed burr, argon beam, phenol and cryotherapy. ABCs in anatomic locations where surgery would cause significant morbidity are most often treated with embolization or radiotherapy and with increasing frequency, medical management with denosumab (RANK-L inhibitor) or percutaneous doxycycline (antibiotic with anti-neoplastic properties) [8, 9]. Current modalities include sclerotherapy, which acts by damaging the endothelium of vessels, triggering the coagulation cascade and resulting in thrombosis [10].

The drawback of sclerotherapy for being unpopular is the risk of local or general inflammatory reactions to the agent and the requirement of curettage and bone grafting at a later point of time.

In a RCT study, Varshney et al. compared Polidocanol (hydroxypolyaethoxydodecan) sclerotherapy with curettage, high speed burr and bone graft; polidocanol had a healing rate of 93.3% compared to 84.8% for curettage. Polidocanol also provided faster pain relief and better functional outcomes [11].

In a study done by Adamsbaum et al., in 2003 on 17 patients with aneurysmal bone cyst treated with sclerotherapy with Ethibloc, 75% patients received a single dose of injection all but one patient developed local and/or general inflammatory reactions a few days after treatment, and three patients had cutaneous fistulas at the site of injection with spontaneous
resolution. Three patients had recurrence (18%), after a mean follow-up of five years; all were treated with surgical resection and bone grafting [12]. In patients sensitive to agents like Ethibloc, sclerotherapy should be avoided and other alternatives like afferent arterial embolization of feeding vessels should be carried out, if the location of the lesion is surgically inaccessible.

In a study done by Brosjö et al., average cumulative dose of polidocanol per patient until healing was 450 (60–1,410) mg. The efficacy of the method was 97% (95% CI: 92–100). 3 injections at intervals of 4 weeks was the most common schedule, and more injections were given if the lesion failed to heal [6].

In our study, we have combined both the conventional method of curettage and cementing with a single sitting sclerotherapy with 3% Polidocanol, followed by plating. Regular follow-ups at 1, 3, 12 and 18 months post-operatively to assess for recurrence of the lesion.

Conclusion
Aneurysmal bone cyst has a higher recurrence rate with curettage and bone grafting alone. Extended curettage along with sclerotherapy with 3% polidocanol has shown no evidence of recurrence and good functional outcome at 18 months follow-up in our study. Hence, this combined approach can be recommended in the management of aneurysmal bone cyst. This study also warrants for a prospective, large sized randomized control trial with a long term follow-up to demonstrate the effectiveness of this approach.

Statement of Consent
Informed and Written consent has been taken from the patient to publish this as a case report for educational purpose.

References
1. Fletcher CD, Bridge JA, Hogendoorn P, Martens F. World Health Organization (WHO) classification of tumours of soft tissue and bone. Pathology and genetics. Lyon: IARC Press 2013;2013:80-2.
2. Park HY, Yang SK, Sheppard WL, Hegde V, Zoller SD, Nelson SD et al. Current management of aneurysmal bone cysts. Current reviews in musculoskeletal medicine 2016;9(4):435-44.
3. Leithner A, Windhager R, Lang S, Haas OA, Kainberger F, Kotz R. Aneurysmal bone cyst. A population based epidemiologic study and literature review. Clinical orthopaedics and related research 1999;1(363):176-9.
4. Martinez V, Sissons HA. Aneurysmal bone cyst. A review of 123 cases including primary lesions and those secondary to other bone pathology. Cancer 1988;61(11):2291-304.
5. Amendola L, Simonetti L, Simoes CE, Bandiera S, De Iure F, Boriani S. Aneurysmal bone cyst of the mobile spine: the therapeutic role of embolization. European Spine Journal 2013;22(3):533-41.
6. Brosjö O, Pechon P, Hasla A, Tsagozis P, Bauer H. Sclerotherapy with polidocanol for treatment of aneurysmal bone cysts: Good results in 37 of 38 consecutive patients. Acta orthopaedica 2013;84(5):502-5.
7. Marcove RC, Sheth DS, Takemoto S, Healey JH. The treatment of aneurysmal bone cyst. Clin Orthop 1995;311:157-163.
8. Dubory A, Missenard G, Domont J. Interest of Denosumab for the treatment of giant-cells tumors and aneurysmal bone cysts of the spine. About nine cases. Spine 2016;41(11):E654-60.
9. Fife RS, Rougraff BT, Proctor C, Sledge Jr GW. Inhibition of proliferation and induction of apoptosis by doxycycline in cultured human osteosarcoma cells. Journal of Laboratory and Clinical Medicine. 1997;130(5):530-4.
10. Falappa P, Fassari FM, Fanelli A, Genovese E, Ascani E, Crostelli M et al. Aneurysmal bone cysts: treatment with direct percutaneous Ethibloc injection. Long-term results. Cardiovascular and interventional radiology 2002;25(4):282-90.
11. Rastogi S, Varshney MK, Trikha V, Khan SA, Choudhury B, Safaya R. Treatment of aneurysmal bone cysts with percutaneous sclerotherapy using polidocanol: a review of 72 cases with long-term follow-up. The Journal of bone and joint surgery. British volume 2006;88(9):1212-6.
12. Adamsbaum C, Mascard E, Guinebretiere JM, Kalifa G, Duboussset J. Intraläsional Ethibloc injections in primary aneurysmal bone cysts: an efficient and safe treatment. Skeletal radiology 2003;32(10):559-66.