

Liquid Absolute Alcohol Based Sclerotherapy - A Boon in Large Grade 3 Aneurysmal Bone Cyst of Proximal Humerus in a Child

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Learning Point of the Article:

96 % Ethanol based intralesional sclerotherapy can be used to manage aggressive ABC lesions in children with favourable outcome.

Abstract

Introduction: Aneurysmal bone cyst (ABC) is a benign intraosseous lesion, usually seen before the age of 20 years and is a lesion filled with blood cavities causing a blowout distension of the bone. It constitutes to about 1% of benign bone tumors. Although benign, a large lesion is liable to develop pathological fracture, so needs prompt addressal. Surgical resection or curettage in large lesions can lead to bone defects, deformities, and even functional abnormalities, especially in children. This article describes a large aggressive ABC of proximal metaphyseodiaphyseal region of proximal humerus in a 12-year-old male patient, which we managed effectively with the use of liquid absolute alcohol based sclerotherapy under fluoroscopic control.

Case Report: A 12-year-old boy presented to the outpatient department of our hospital presenting with complaint of swelling in the right shoulder region which was insidious in onset with gradual increase in size and deep aching pain since past 6 months. Plain radiograph revealed a large expansile osteolytic lesion with characteristic blown out “soap bubble appearance” involving the proximal humerus and abutting the growth plate. Hence, percutaneous needle biopsy of the lesion under fluoroscopic guidance was undertaken. The histology was likened to a “blood-filled sponge” composed of blood-filled anastomosing cystic cavernomatous spaces separated by wall composed of fibroblasts, myofibroblasts, and osteoclast such as giant cells, osteoid, and woven bone confirmed the diagnosis of ABC. Radiologically, it was classified as Enneking Stage 3 [1] cyst which is locally aggressive and expanding with significant cortical destruction and Capanna type 2 [2] lesion involving the entire bony segment (proximal metaphyseodiaphyseal region) with marked expansion and cortical thinning. Although resection/excision or curettage with bone grafting are commonly undertaken, concerns were for issues of subsequent bony reconstruction given the size of defect with possibility of need of an implant for stabilization, likelihood of damage to growth plate and functional compromise the shoulder. Hence, a decision to treat the patient with liquid absolute alcohol based sclerotherapy was planned.

Conclusion: Sclerotherapy with ethanol 96% is a useful method for the treatment of large aggressive ABC, especially in children. It is a minimally invasive method, with no major complications, which lowers the risks of open surgical intervention and has a good outcome when undertaken with proper precautions.

Keywords: Aneurysmal bone cyst, pediatric bone tumors, bone cysts, absolute alcohol, sclerotherapy.

Introduction

Aneurysmal bone cyst (ABC) is a benign tumor affecting the skeletal system and occurring usually during growth period. It is locally destructive to the bone. Surgical resection or curettage in large lesions can lead to bone defects, deformities, and even functional abnormalities, especially in children. This article

describes a large aggressive ABC of proximal metaphyseodiaphyseal region of proximal humerus in a 12-year-old male patient who had a complaint of slowly increasing swelling with aching pain in the right shoulder region with no other associated problems. In this case, we used three sessions (decided on the basis of response of sclerotherapy during

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Author's Photo Gallery



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Figure 1: Radiograph of proximal humerus showing Grade 3 soap bubble like cystic lesion involving metaphysis and upper diaphysis with good shoulder abduction.

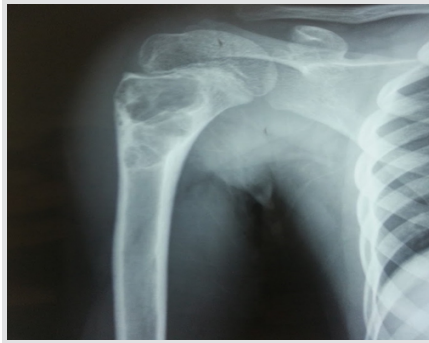


Figure 2: Radiograph of proximal humerus showing Grade 3 capanna Stage 2 lesion proximal humerus with soap bubble appearance likened to be aneurysmal bone cyst.

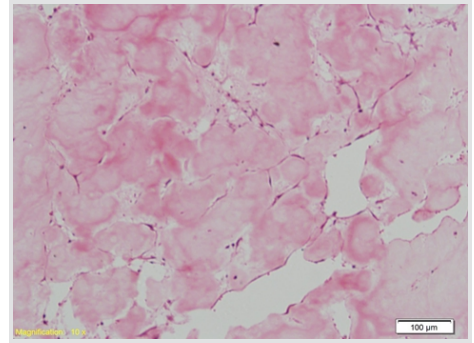


Figure 3: Histopathology showing anastomosing blood filled cavernomatous spaces walled with fibroblasts-like a blood filled sponge appearance.

follow-up which was made at 3 monthly interval) of percutaneous sclerotherapy with absolute alcohol as an sclerosing agent, which is easily available in the hospitals under fluoroscopy avoiding surgical resection or curettage due to large size of the tumor and its proximity to growth plate. When analyzed on follow-up imaging, bone defect got reconstituted with resolution of the lesion without compromising growth plate or shoulder function after intralesional sclerotherapy with absolute alcohol. We have not found the successful use of absolute alcohol for management of large Enneking Stage 3 [1] and Capanna type 2 lesion of proximal humerus in a young child of 12 years. Thus, use of absolute alcohol is a good feasible alternative treatment for large ABC lesions, especially in children just adjacent to the growth plate, where the traditionally described method of curettage and cementing or bone grafting may be associated with significant morbidity due to its possibility of causing damage to growth plate, need of large volume of bone graft, and need of additional support with implant in the weakened bone area following surgical resection or curettage.

Case Report

A 12-year-old boy presented to the outpatient department of our hospital presenting with complaint of swelling in the right shoulder region which was insidious in onset with gradual increase in size and deep aching pain since past 6 months. Patient had no history of trauma, any constitutional or systemic

complaints. On local examination, there was diffuse swelling involving the right shoulder region with slight increase in local temperature and tenderness of proximal humerus. The movement around the shoulder was painful and there was no associated neurovascular deficit in the upper limb. Patient was subjected to radiograph of the proximal shoulder. All the hematological and routine biochemical parameters were normal except for mild increase in erythrocyte sedimentation rate (ESR-32 mm/1st h). Plain radiograph revealed a large expansile osteolytic lesion with characteristic blown out “soap bubble appearance” (Fig. 1, 2) involving the proximal humerus and abutting the growth plate.

Radiologically it was classified as Enneking Stage 3 [1] cyst which is locally aggressive and expanding with significant cortical destruction and Capanna type 2 [2] lesion involving the entire bony segment (proximal metaphyseodiaphyseal region) with marked expansion and cortical thinning. Usually, clinical and radiologic features may be sufficient for a presumptive diagnosis of ABC, but establishing the histological diagnosis is essential before committing to definitive treatment. Hence, percutaneous needle biopsy of the lesion under fluoroscopic guidance was undertaken. The histology was likened to a “blood-filled sponge” composed of blood-filled anastomosing cystic cavernomatous spaces separated by wall composed of fibroblasts, myofibroblasts, and osteoclast such as giant cells, osteoid, and woven bone [3, 4, 5] confirmed the diagnosis of ABC (Fig. 3, 4, 5). Due to the unusually large bony involvement, osteolytic and expansile characteristic of the

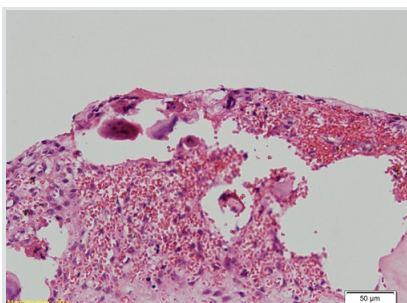


Figure 4: Histological slide showing apart from cavernous RBC filled spaces and interspersed giant cells with fibroblastic cells lining the cysts.

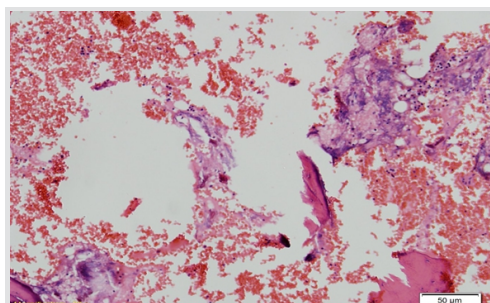


Figure 5: Microscopic examination revealing apart from RBC cavernomatous cystic spaces with poorly interspersed trabeculae with osteoblasts, likened it to be aneurysmal bone cyst.



Figure 6: Radiograph showing the lesion getting calcified and bone getting remodeled after 2nd intralesional injection of liquid absolute alcohol at 6 months with intact growth plate.

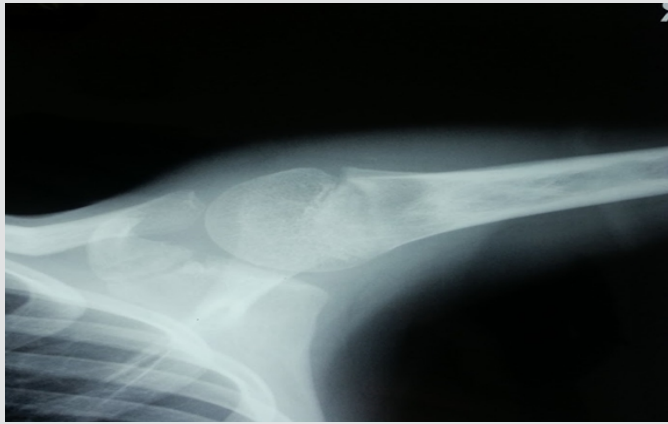


Figure 7: Radiograph showing the lesion getting completely ossified with remodeled bone with good shoulder abduction and intact growth plate at 9 months (3 months after the third injection of liquid absolute alcohol).

lesion intervention is preferred over mere observation. Although resection/excision or curettage with bone grafting is commonly undertaken, concerns were for issues of subsequent bony reconstruction given the size of defect with possibility of need of an implant for stabilization, likelihood of damage to growth plate and functional compromise of the shoulder. Hence, a decision to treat the patient with liquid absolute alcohol based sclerotherapy was planned. This being a harmless and easily available material in most hospital settings, while other agents which have been tried for sclerotherapy may not be easily available. Sclerotherapy was done under general anesthesia with fluoroscopic guidance to ensure correct positioning of 18 G needle into the lesion. We proceeded to injecting 96% ethanol in the dosage of 1 ml/kg. Higher doses of alcohol in children can lead to alcohol poisoning with severe symptoms [6]. Moreover, precaution was taken to stop drug administration when increased resistance to injection was observed to prevent extravasation into especially draining veins and surrounding soft tissues and other drug related complications. After 10 min, the alcohol was subsequently flushed with saline. Following the procedure, the patient was given nonsteroidal anti-inflammatory drugs (Tab. Ibuprofen 400 mg twice a day) for 3 days with a 5 days course of broad spectrum antibiotics (Tab. Cefuroxime 250 mg twice a day), to minimize pain and local inflammatory reaction that may follow the injection. Patient was hospitalized for 48 h especially to monitor injection associated complications such as local inflammatory reaction, severe pain, and dizziness after injection. The radiological follow-up was performed after 3 months and based on the radiological response by observing reossification of the lesion, the therapy was repeated at 3 and 6 months follow-up. Patient showed almost complete reconstitution of lesion and bone remodeling with good shoulder function and intact growth plate (Fig. 6, 7). In search of literature, we have not found such a large sized aggressive ABC of proximal humerus at an early age of 12 years, managed

successfully with a commonly available sclerotherapy agent, that is, absolute alcohol [7].

Discussion

ABC is a benign intraosseous lesion, usually seen before the age of 20 years and is a lesion filled with blood cavities causing a blowout distension of the bone. It constitutes to about 1% of benign bone tumors. Although benign, a large lesion is liable to develop pathological fracture, so needs prompt addressal. Percutaneous needle biopsy, with or without image guidance (fluoroscopy/ultrasound/computed tomography) depending upon location, is considered the preferred method for definitive diagnosis. Treatment range from non-invasive to invasive involving en bloc surgical excision, intralesional curettage with or without local adjuvants, and minimal invasive surgical techniques such as embolization, sclerotherapy, and radiotherapy [8]. At present, trend is to use sclerotherapy with trials of wide range of sclerotherapy agents. Intralesional injection of drugs is preferred because it offers a least invasive therapeutic option. It works by causing damage to vascular endothelium initiating the cascade of events resulting in healing of the lesion. Various agents have been described for sclerotherapy such as percutaneous intralesional administration of ethanol 96%, Aethoxysklerol 3% (polidocanol- hydroxypolyaethoxydodecan) [9], Ethibloc (a hydroalcoholic radiopaque solution of zein) [10], triamcinolone acetonide [11], liquid absolute alcohol, and absolute alcohol gel [12, 13]. Intravenous denosumab [14] has been tried for ABCs in anatomically critical locations. Image guided percutaneous cryoablation and embolization with N-2-butyl-cyanoacrylate have been used successfully in cases of spinal ABC [15]. Studies on percutaneous injection of concentrated stem cells from bone marrow [16] and injection of whole bone marrow have shown to be effective in cyst resolution [17, 18], presumably through bone marrow derived stem cell differentiation into osteoblastic lineage leading to new bone formation. Absolute alcohol is a very good sclerotherapy agent in ABC and its use under fluoroscopy contributes to limiting complications in sclerotherapy, by providing instantaneous visualization of diffusion, enabling injection to be stopped in case of extravasation. Although the optimal efficient method of treatment for ABC is still unclear especially with regard to percutaneous sclerotherapy agents [19], absolute alcohol may be considered with its easy availability and harmless nature as seen in our case, especially if used properly.

Conclusion

Sclerotherapy with ethanol 96% is a useful method for the treatment of large aggressive ABC, especially in children. It is a

minimally invasive method, with no major complications, which lowers the risks of open surgical intervention and has a good outcome when undertaken with proper precautions. However, more large randomized controlled trials may be required with the use of various sclerosing agents to find their relative clinical efficacy and recommendations.

Clinical Message

Liquid absolute alcohol can be effectively tried in pediatric patients of large aggressive ABCs, especially when it is easily available in most hospitals and has presumably good outcome.

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