A Rare Case of Aneurysmal Bone Cyst – Navicular Bone Treated by Intralesional Sclerotherapy Agent Polidocanol

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Learning Point of the Article:
Role of Sclerotherapy in the management of aneurysmal bone cyst and it's advantages over conventional surgical curettage in smaller lesions.

Abstract

Introduction: Aneurysmal bone cyst (ABC) is an intraosseous arteriovenous malformation, usually treated by surgical excision but with higher recurrence rates. The occurrence of this lesion in navicular bone is a rare entity. Repetitive sclerotherapy using 3% polidocanol is a minimally invasive, safer method of treatment for ABCs.

Methods: A patient of ABC of the navicular bone precisely diagnosed through X-ray, MRI, and biopsy was given periodic injections of 4 ml of 3% polidocanol under aseptic precautions from April 2018 to July 2018. A total of four injections given at an interval of 1 month.

Results: Regular follow-up done for 1 year. At the end of all the four injections, the lesion healed completely with a relief in presenting symptoms. No complications were reported. At 3 months post-therapy, the patient was able to walk without pain. After 1 year of follow-up, the patient was symptom-free with no signs of recurrence.

Conclusion: Surgical resection/curettage has higher chances of recurrence; therefore, usage of intralesional sclerotherapy agent polidocanol should be considered the treatment of choice in the lesions smaller than 5 cm.

Keywords: Aneurysmal bone cyst, sclerotherapy, polidocanol.

Introduction

An aneurysmal bone cyst (ABC) is neither a cyst nor a neoplasm. The current theory is that the lesion is an intraosseous arteriovenous malformation [1]. Thus, it is a benign blood-filled lesion with a cyst-like wall predominantly of fibrous tissue and surrounded by a thin layer of periosteal new bone [2, 3, 4, 5]. The foot is a relatively uncommon location for an ABC. To the best of our knowledge, no previous case of ABC has been reported in the navicular bone. In the case reported here, an extensive lesion in the navicular was treated with an intralesional sclerotherapy agent polidocanol. Repetitive sclerotherapy using polidocanol is a minimally invasive, safer method of treatment for ABCs as compared with intralesional excision and bone grafting [6].

An 18-year-old man in otherwise good health presented complaining of the right midfoot pain and swelling for 7 months, worsening over the preceding 3 months. He stated that he could walk, but this was associated with pain. A 2 × 3 cm firm, tender mass was palpated on the medial aspect of his right midfoot (Fig. 1).

He had full range of motion at his ankle and limited inversion and eversion secondary to pain. A roentgenogram (Fig. 2) showed an expansile, lytic lesion in the navicular bone. Magnetic resonance imaging (Fig. 3-5) shows altered signal intensity over navicular bone of size 2.1 × 2.8 cm with mild edema and infiltration over surrounding soft tissues.

No other associated lesions were seen. Under strict aseptic precautions, biopsy was taken, it was a thick column of blood
Pain and/or swelling are the most common symptoms of an ABC on presentation [7]. Patients notice this increase in size due to both the lesion increasing in size as well as associated swelling. The vast majority (~80%) of patients with primary ABCs are younger than 20 years old. Women are affected slightly more often than men. ABC shows an evident predilection for the vertebral column and long bones – particularly the femur, humerus, tibia, and fibula [8]. The feet account for approximately 4% of ABCs [9]. The natural history is characterized by a benign lesion that does not metastasize yet can be difficult to eradicate with simple curettage. The greatest clinical problem associated with the ABCs is local recurrence. As reported for two large series, the factors associated with a higher recurrence rate in ABC seemed to be young age and/or the size of the cyst. In addition, Tillman et al. noted that cysts localized in the major limb bones had a higher recurrence rate. The cause of ABCs remains unclear, although several theories have been suggested. One theory by Lichtenstein proposes that the lesion originates as a circulatory disturbance, for instance, in response to increased intraosseous venous pressure. This engorged vascular bed then causes resorption of spongy bone, erosion of cortical bone, and expansion of the lesion. A second theory proposes that the cyst appears as a dilated arteriovenous shunt. A third theory by Bernier and Bhaskar proposes that the ABC begins as a focus of intramedullary hemorrhage and that the lesion represents an unusual reparative response to hemorrhage. The fourth theory

Discussion

Pain and/or swelling are the most common symptoms of an aspirate, and results (Fig. 6) were consistent with ABC. In view of high recurrence rate after curettage of ABC, the primary procedure of intraleseional sclerotherapy was planned. Under the aseptic condition, hypodermic needle inserted into the lesion with the help of image intensifier, aspirated to confirm the site followed by intraleseional injection of polidocanol, a sclerotherapy agent at a dosage of 2 mg/kg, with the patient being 60 kg a total of 120 mg, was given. It was performed as a daycare procedure and carried out under local anesthesia. Early rehabilitation was performed with no mode of immobilization. Four such doses were given periodically at an interval of 1 month under aseptic precautions using the same aspiration and injection technique. The response was regularly observed with roentgenograms in each visit. The outcome seems to be markedly appreciable since the first injection and at the end of all the four injections, lesion found to be remitted completely and the patient improved greatly in view of symptoms. No complications were reported. At 3 months post-therapy (Fig. 7, 8), roentgenogram was done and the patient was able to walk without pain and range of movements was full and painless in ankle and subtalar joint. The patient is symptom-free and shows no signs of recurrence at 1-year follow-up (Fig. 9).

Figure 1: Clinical photograph showing swelling over medial aspect of the right foot.

Figure 2: X-ray showing expansile lytic lesion in the navicular bone.

Figure 3: MRI image showing altered signal intensity over navicular bone.

Figure 4: MRI image showing altered signal intensity over navicular bone.

Figure 5: MRI report.

Figure 6: Biopsy report.

Figure 7: X-ray 3 months post-therapy, showing sclerosed areas in the navicular bone.

Figure 8: Follow-up clinical image 3 months after therapy.
A definitive diagnosis of ABC can be made only by its microscopic features. The lesion is a pseudocyst composed of blood pools of variable sizes, lined not by endothelium but by spindle-shaped cells and multinucleated giant cells. The stroma is composed of giant cells, fibroblasts, capillaries, extravasated blood cells, and hemosiderin. The walls of the blood pools do not contain elastic elements or smooth muscle and, thus, do not mimic blood vessels.

Before any therapy can be considered, it must be determined whether the ABC is associated with another condition. If a more aggressive lesion is present, treatment must be directed toward the more aggressive component. Conventional treatment has been directed toward the surgical removal of the entire lesion, or of as much as possible, followed by packing with bone graft. This common method of treatment is usually curative. Incomplete surgical removal is the most important factor in recurrence. Recurrence rates range from 10% to 44%, with 90% recurring within 2 years [5, 7, 10, 15]. Sequential percutaneous administration of polidocanol is a safe, simple procedure with an excellent cure rate for ABC. The use of polidocanol is a definite advancement over previous sclerotherapy regimens that relied on alcoholic zein solutions, which were more toxic and had serious adverse effects after spill-out into nearby tissues [16, 17]. Being a daycare procedure of negligible risk of morbidity and no scar formation with efficacy exceeding 90%, it outweighs the disadvantage of multiple injections and prolonged treatment.

**Conclusion**

Surgical resection/curettage has higher chances of recurrence, and so usage of intralesional sclerotherapy agent polidocanol should be considered to be the treatment of choice in the lesions smaller than 5 cm, as was the case with the patient described in this report.

**Clinical Message**

Sclerotherapy possesses a great advantage of being minimally invasive technique and with less recurrence rates. Being a daycare procedure with early rehabilitation, it offers a comfortable and compliant mode of therapy for the patients.

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