

Case Report

Minimally invasive technique for recurrence of Aneurysmal bone cyst following extensive operative procedure- A case report

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Abstract: A 3 year old girl diagnosed with pathological fracture of the lower end of right tibia due to Aneurysmal Bone Cyst (histopathological and radiological diagnosis) who underwent curettage of the lytic lesion and filling of the defect with fibula strut graft and synthetic bone substitute, presented to us three months after the procedure due to recurrence of pain and limp. X-ray shows recurrence of radiolucency at the distal end of tibia. The location and dimension of the lesion was ascertained with a CT scan of the leg. With a bone biopsy needle, contents aspirated and filled with 3% Polidocanol which was repeated after a 4 week interval. She was evaluated periodically with radiographs. She was asymptomatic and started playful activities with ease. Each successive radiograph showed regression of the lytic lesion and reduction of pain. Last X-ray taken one and half years post infiltration showed no evidence of a lytic lesion.

Keywords: Aneurysmal bone cyst; recurrent; Sclerosants; polidocanol

INTRODUCTION

The aneurysmal bone cyst (ABC) is a benign solitary osteolytic and expansile bone tumor. It is extremely rare, occurring in 1.5 per one million persons per year [1]. It most commonly occurs in the metaphysis of long bones during the first two decades of life and usually presents due to pain, swelling, or pathological fracture [2]. The etiology of ABC is unknown but numerous authors have proposed that it forms in response to vascular disruption in the bone due to a preexisting primary tumor or a traumatic insult [3-7].

They usually manifest as expansile osteolytic lesions with a varying potential to be locally aggressive. Since their first description in 1942, a variety of treatment methods has been proposed. Traditionally, these tumors were treated with open surgery. Either intralesional surgical procedures or *en bloc* excisions have been described. Furthermore, a variety of chemical or physical adjuvants have been utilized in order to reduce the risk for local recurrence after excision. Currently, there is a shift to more minimally invasive procedures in order to avoid the complications of open surgical excision. Good results have been reported during percutaneous surgery, or the use of embolization. Recently, sclerotherapy has emerged as a

promising treatment, showing effective consolidation of the lesions and functional results that appear to be superior to the ones of open surgery [8].

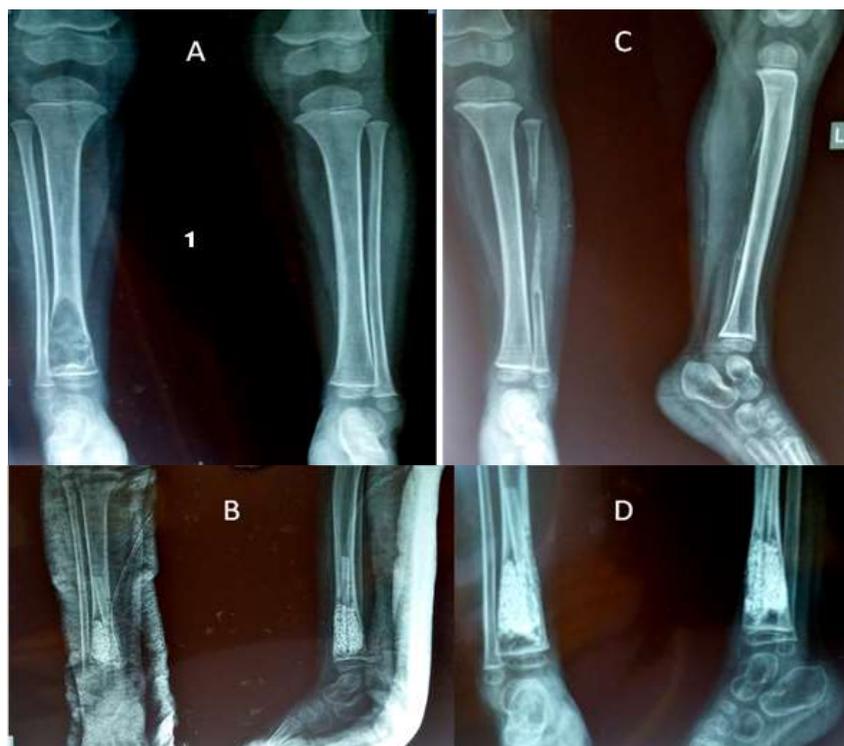
CASE REPORT

A three year old girl presented with her parents to our institution with pain around the right ankle joint region and limp two years ago. On discussion with her parents and detailed clinical evaluation it was concluded that she was diagnosed to have aneurysmal bone cyst (Radiologically and Histologically) at the distal 1/3 region of the right tibia. She had undergone fibula strut graft from the contralateral leg and synthetic bone substitution, following curettage of the lytic region two years and three months ago (Figure 1). She was kept non-weight bearing for 5 weeks. She developed pain 2 months after the surgery and came to our institution. Radiological evaluation showed partial incorporation of the graft and a recurrence of lytic lesion in the distal portion of the right tibia. She was advised for regular follow-ups with x-rays, but her next review after 5 months she presented with worsening of signs and symptoms. X-rays showed progression of the lytic lesion. A CT scan of the right distal tibia and ankle was done, which showed features of recurrent lesion involving about half of the distal portion of tibia, sparing the epiphysis (Figure 2).

A plan was made to infiltrate the lesion with sclerosing agent. An 18G bone biopsy needle was used, and introduced into the area of recurrence. Aspiration was done and 1ml of 3% polidocanol was infiltrated. Back flow was prevented by locking the needle for 1 minute and then was flushed with 0.5ml of normal saline. This was done to prevent extravasation of the polidocanol and to prevent any skin hypopigmentation (Figure 3). The same procedure was repeated after 4 weeks as there was not much change in the radiographs (Figure 4). X-ray was repeated two months later. The

fibula strut graft and the synthetic bone graft were being incorporated and the area of radiolucency was decreasing in size. Regular follow-ups were done and the patient was feeling better.

Six months after the sclerotherapy, the patient was completely asymptomatic. Radiographs showed complete incorporation of the graft and the lytic lesion was barely visible. She was followed up for one and half years till august 2016. Recent X-rays shows no evidence of recurrence (Figure 5).



**Fig-1: a) Pathological fracture at the right tibia distal aspect
b) Immediate post-fibular strut graft and synthetic bone graft
c) Contralateral leg showing graft donor site
d) Post-op x-rays showing partial incorporation of graft.**



Fig-2: X-ray and CT scan showing recurrence of lesion.



Fig-3:Aspiration of cavity and infiltration of sclerosing agent, post-op X-ray.



Fig-4: Before and after second infiltration of sclerosant.



Fig-5: Latest X-ray, one and a half years post infiltration.

DISCUSSION

Aneurysmal bone cyst is a rare benign bone tumor that contains blood-filled cavernous spaces separated by septa containing osteoid tissue and osteoclast giant cells. Young patients are most often affected with tumors located at the metaphysis of long bones. In less frequent occasions aneurysmal bone cysts found in pelvis and spine, and sometimes aneurysmal bone cyst could be present with aggressive behavior [9-11]. Aneurysmal bone cyst can be present early before reaching giant size, which facilitates early diagnosis and treatment. It is well known that Aneurysmal bone cyst is classified as an aggressive benign bone tumor, which means that if not treated properly, it may recur or if left untreated, it may get larger and eventually grow to be a giant Aneurysmal bone cyst [12]. These aggressive lesions are difficult to address and could be challenging to any orthopaedic surgeon.

Although the pathogenesis of Aneurysmal bone cyst is still unknown, they can be considered either primary (70%) or secondary (30%) [13].

Primary Aneurysmal bone cysts arise *de novo*. A secondary Aneurysmal bone cyst develops in association with other neoplasms most commonly giant bone tumor (GCT) of the bone, osteoblastoma, chondroblastoma, and fibrous dysplasia [14].

Radiographically, the diagnosis of an Aneurysmal bone cyst shows five classic findings [11]. First, the neoplasm is typically an expansile lytic lesion with a soap-bubble appearance. Second, it presents as an eccentric lesion outlined by a thin layer of subperiosteal new bone. Third, it presents as a centric lesion. Fourth, it is a metaphyseal lesion that occupies a large percentage of the bone with trabeculations at the edges. Fifth, it results in soft tissue expansion and destruction of the cortex. Additionally, it is suggested that if the cyst's transverse diameter on radiographic examination is equal to or more than three times the diameter of the adjacent normal bone, it can be called giant Aneurysmal bone cyst [13].

Many forms of treatment have been described such as intralesional procedures, radiation therapy, subtotal or total excision with or without reconstruction, sclerotherapy and selective/super-selective embolisation [9, 14-18]. It has also been reported to regress spontaneously after biopsy and fracture healing. Curettage with or without bone grafting is considered to be the treatment of choice for these lesions. It has also been reported to regress spontaneously [19] after biopsy and fracture healing, however, a high recurrence rate and the potential of disturbance of growth when the lesion is near a physal plate [9,14-16]. Marcove *et al.* [14] and Cole [16] reported recurrence rates of more than 50% after curettage with or without bone grafting.

Schreuder *et al.* [17] reviewed the literature and found a recurrence rate of 30.8% after curettage and bone grafting. High-speed burring and cryotherapy [14, 17] have been used as adjuvants in an attempt to reduce the recurrence rate, to as low as 4% [17]. However, these procedures are associated with haemorrhage, particularly in large lesions, incomplete excision and physal injury [14,16,20]. Moreover, surgery is not feasible at all sites due to anatomical constraints. Endoscopic curettage has also been described in a small series of patients but long-term results are awaited [18]. Although extralesional excision removes the lesion *in toto*, it may involve extensive surgery with prolonged immobilization, and may be complicated by the need for bone grafts and associated, growth plate injury and significant morbidity [21]. Saucerisation of the lesion has similar complications although complete healing has been seen [14,16].

Radiotherapy has now been abandoned in view of the large number of complications including malignant change. Marcove *et al.* reported uncontrolled lesions in 9% of their patients and the development of a secondary tumour in one patient after radiotherapy [9,14,21].

Embolisation was used to reduce the intra-operative blood loss and proved to be an effective method of treatment. A recurrence rate of 10.5% was reported by De Cristofaro *et al.*, Super selective embolisation of the feeding vessels widened the application of this method to surgically-inaccessible regions. Although the procedure is very effective, not all aneurysmal bone cysts have a major feeding vessel which can be satisfactorily embolised. Also ischaemia of vital neural and visceral structures remains a major concern [22, 23].

Sclerosants, in general, act by direct damage to the endothelial lining, triggering a coagulation cascade and thrombotic occlusion of blood vessels [24, 25]. Several sclerosing agents have been used, an alcoholic solution of Zein [26-29] is the most popular.

Polidocanol has been used safely and with efficacy in the treatment of vascular malformations at various sites with relatively few complications [24,25]. Methylprednisolone acetate and radionucleides have also been used sporadically [30].

Polidocanol sclerotherapy has an efficacy exceeding 90%. Furthermore, the treatment is simple and carries negligible risk of morbidity, there is no scar formation, and it can be performed as a day-case surgery. The method is applicable to all cases, and does not require sophisticated equipment. Most importantly, sclerotherapy is effective in the case of lesions of the pelvis and sacrum that are difficult to treat surgically

due to the risk of heavy bleeding and other major complications [31,32].

The obvious disadvantage is the need for multiple injections [33].

The use of polidocanol sclerotherapy 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 [26].

CONCLUSION

Once histopathological and radiological confirmation of Aneurysmal bone cyst is done, sclerotherapy procedure is an ideal mode of treatment. It is repeatable with almost nil adverse reactions. It is minimally invasive, cost effective. Polidocanol has efficacy exceeding 90% and has a proven track record.

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