

## Effectiveness of Sclerotherapy in Treatment of Patients with Primary Aneurysmal Bone Cysts at King Abdulaziz Medical City (KAMC)

Wazzan ALjuhani<sup>1</sup>, Badr Almana<sup>2\*</sup> and Mohammed Alotaibi<sup>3</sup>

<sup>1</sup>Assistant Professor, King Saud Bin Abdulaziz University for Health Science, King Abdullah International Medical Research Center, King Abdulaziz Medical City Surgery Department, Riyadh, Saudi Arabia

<sup>2</sup>Medical Student at King Saud bin Abdulaziz University for Health Sciences, Riyadh, Saudi Arabia

<sup>3</sup>Deputy Chairman for KASCH, Consultant Pediatric Radiologist and Interventional Radiologist, King Abdullah Specialist Children's Hospital, Riyadh, Saudi Arabia

\*Corresponding Author: Badr Almana, Medical Student at King Saud bin Abdulaziz University for health sciences, Riyadh, Saudi Arabia.

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### Abstract

**Introduction:** Aneurysmal bone cysts (ABC) are benign expansile osteolytic tumor-like lesions that are characterized by multiple blood-filled cavities [1]. They treated by percutaneous sclerotherapy is done by an interventional radiologist, and the procedural method is completed by injecting a sclerosing agent (sodium tetradecyl sulfate, doxycycline, ethibloc, ...etc) into the lesion under radiological guidance [10,11].

**Material and Methods:** A single-center retrospective study covered the period 2008 – 2019. Nine pediatric patients with primary ABC managed by sclerotherapy were included. The most used fibrosing agent in this study is doxycycline, followed by sodium tetradecyl sulfate, then polidocanol.

**Results:** Our analysis showed that five patients had excellent response (55.55%) with more than 50% ossification. Only one patient (11.11%) had poor response with less than 25% ossification. Three patients had partial response based on radiological evaluation with 25% - 50% ossification.

**Discussion:** Despite the small sample size of our study due to rarity of the disease and sporadic nature of the ABC, though it showed an efficacy and safety of the sclerotherapy which is a minimally invasive procedure done under image guidance.

**Keywords:** Sclerotherapy; Aneurysmal Bone Cyst; Polidocanol; Doxycycline; STS

### Introduction

Aneurysmal bone cysts (ABC) are benign expansile osteolytic tumor-like lesions that are characterized by multiple blood-filled cavities [1]. It can affect any bone in the body, but most commonly in long bones especially in the eccentric metaphysis [2]. The clinical presentation is mainly pain, swelling, and possible deformity due its ability to expand which may lead to pathological fracture, and restriction of movement [3]. It may also cause neurologic symptoms (location dependent) [4]. The exact etiology of ABC is unknown. However, most researchers believe that it is due to in-

creased venous pressure or malformation. ABC could be primary, or secondary to another underlying lesion like osteoblastoma, osteosarcoma, fibrous dysplasia, ...etc. Recent studies have found that 69% of primary ABC are due to t (16;17) translocation which leads to upregulation TRE17/USP6 oncogene. However, secondary ABCs do not explain genetic changes [5-7]. Aneurysmal bone cysts are rare, and most cases are within the pediatric groups [8]. It can be treated by curettage and bone grafting with approximately 20% - 25% recurrence rate [9]. A minimally invasive therapy is also performed percutaneously (sclerotherapy) [10,11].

Aneurysmal bone cysts treated by percutaneous sclerotherapy is done by an interventional radiologist, and the procedural method is completed by injecting a sclerosing agent (sodium tetradecyl sulfate, doxycycline, ethibloc, ...etc) into the lesion under radiological guidance [10,11]. Many studies have shown that sclerotherapy is good and minimally invasive alternative method when it comes to treatment of aneurysmal bone cysts. One study showed complete ossification in the majority of its population and 94.7% were pain-free after 3 months [12]. Another study have shown that using a certain fibrosing agent (i.e doxycycline) in treatment of ABCs is associated with great healing response and a recurrence rate of 5% at more than 2 years [13]. A study showed excellent regression in the lesion's size in 94% of the chosen population [14]. Moreover, a published study was done on 38 consecutive patients showed that sclerotherapy with polidocanol has great efficacy in treating ABCs and is a safe alternative way to conventional surgery, especially those who are difficult to manage, or associated with higher morbidity [15]. There is also improvement in the functional score, with high reduction in the lesion's size when it comes to treatment of ABCs by sclerotherapy [16].

## Materials and Methods

A single-center retrospective study covered the period 2008 – 2019. Nine paediatric patients with primary ABC managed by sclerotherapy were included. All consents for the procedure were taken from the child's parents or legal guardian. Adults and all secondary ABC patients were considered as exclusion criteria. Out of the nine patients, five were males. The mean age, 10 years (range: 4 –16 years). Two patients presented with pathological fracture secondary to ABC. Five of total nine patients are still on follow-up (55.55%), three patients lost follow-up, and one patient with a follow-up of 3 years.

The variables used during data collection were: gender, age of onset, location of the cyst, circumstances of diagnosis, number of sclerotherapy sessions, sclerosing agent, preoperative and post-operative size, duration of healing, complications, follow-up, and most importantly, the outcome. Preoperative and postoperative evaluation of the cyst included both plain radiographs and magnetic resonance imaging. Postoperative evaluations were done 6 months apart after the sclerotherapy session. Ossification of more than 50% is considered excellent response, 25% - 50% is considered partial response, less than 25% is considered as poor response. The procedure is done under general anaesthesia. The

most used fibrosing agent in this study is doxycycline, followed by sodium tetradecyl sul fate, then polidocanol. Re-injection is recommended when there is poor or partial response after 6 months follow-up from the 1<sup>st</sup> session of sclerotherapy.

## Results

Our analysis showed that five patients had excellent response (55.55%) with more than 50% ossification, all of which are currently pain-free. Three of these patients are still on follow up, one with 3 years follow-up, and one lost follow up. Only one patient (11.11%) had poor response with less than 25% ossification lost follow-up, as a result, we were not able to asses any further healing process. Three out of nine patients had partial response based on radiological evaluation with 25% - 50% ossification, two of which are still on follow-up, and one lost follow-up. There were no complications in all patients. Eight patients had ABC in their long bones, whereas only one patient presented with sacral ABC and underwent 4 injections with partial response but clinically improved and pain-free. Among the eight patients who presented with long bone ABC, only two had the cyst in the distal part. Patient number 4 and number 8 presented with pathological fracture secondary the ABC and all of which were in the proximal femur. Regarding of the circumstances of diagnosis, the only complains were swelling, and pain, with no accidental findings. However, 7 patients complained of pain with or without swelling, and only two presented with swelling without pain. The mean of sclerotherapy sessions was 2, only one patient underwent 4 sclerotherapy sessions with partial response in the sacrum. Doxycycline was injected in seven patients, only 57% of injected doxycycline had an excellent outcome, and 43% had a partial outcome, and no poor responses. STS were injected in four patients, two with excellent responses, one partial, and one poor response. Polidocanol was administered in only 2 patients, once in patient number 8 with excellent response, and once in the second sclerotherapy session is patient number 9 with partial response. Three patients had total resolution on radiological evaluation, and increased post-operative size in patient 5 who lost the follow-up. In patient 7, the ABC enlarged by 2 cm based on plain radiograph after the first sclerotherapy session, but ended with excellent response after the second sessions. The mean of duration of healing is 20 months. The only patient who had poor response with an increase of the cyst size after the sclerotherapy sessions had duration of healing of 5 years.

Patient	Gender	Age of onset	Location	Circumstances of diagnosis	Sclerotherapy sessions	Sclerosing agent	Pre-operative size	Post-operative size	Duration of healing	Outcome	Complications	Follow-up
1	F	8	4th metatarsal	Pain	3	35 mg doxycycline	1.7x3.1 cm	Total resolution	3 years	Excellent	None	Still on follow-up
2	M	8	Distal ulna	Pain, swelling	1	35 mg doxycycline	6x8 cm	4.1x6 cm	3 months	Partial	None	Lost follow-up
3	F	8	Proximal fibula	Pain	1	100 mg doxycycline	2.8x3.7 cm	2.2x2.6 cm	5 months	Excellent	None	Lost follow-up
4	M	2	Proximal femur	Pain	3	30 mg doxycycline	2x3 cm	1.4x1.5 cm	1.5 years	Partial	None	Still on follow-up
5	M	2	Proximal femur	Swelling	2	3% STS	3.2x3.3 cm	7.8x4.1 cm	5 years	Poor	None	Lost follow-up
6	F	11	Proximal fibula	Pain, swelling	1	3% STS	3x1.1 cm	Total resolution	8 months	Excellent	None	3 years
7	M	2	Proximal tibia	Swelling	2	100 mg doxycycline - 300 mg doxycycline	4.4x4 cm	Total resolution	1.5 years	Excellent	None	Still on follow-up
8	M	5	Proximal femur	Pain	2	1% STS 3% polidocanol	4x3.6 cm	1.6x1.7 cm	1.5 years	Excellent	None	Still on follow-up
9	F	9	Sacrum	Pain, swelling	4	200 mg doxycycline - 93% polidocanol - 3% STS - 70 mg doxycycline	8.1x7.1 cm	5.6x7.2 cm	1.5 years	Partial	None	Still on follow-up

Table 1

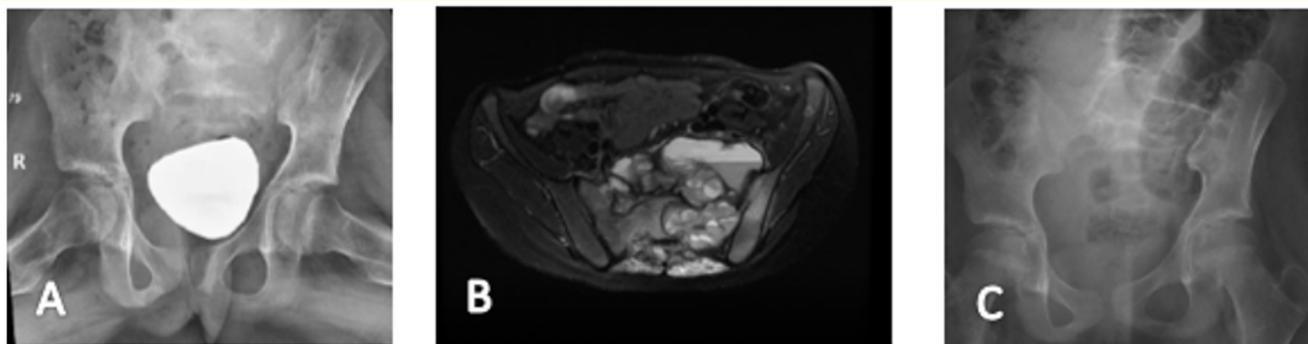
## Discussion

Our study demonstrated and proved the benefits of minimally invasive treatment for ABC. Pain was the most presenting symptoms (78%) followed by swelling (56%). Pain is the most frequent revealing factor in previous studies as was the case in the present study [17,18]. Despite the small sample size of our study due to rarity of the disease and sporadic nature of the ABC, though it showed an efficacy and safety of the sclerotherapy which is a minimally invasive procedure done under radiological guidance. Our study showed that eight patients (89%) had either excellent or good response and only one patient (11%) demonstrated poor response which is hypothetically attributed to high blood flow within the cyst evident by rapid washout of the contrast from the lesion after cystography. The sclerosant needs to have maximum contact time to the wall of

the cysts and if there is rapid washout of the contrast prior to injection this will be considered a poor predictor for the cyst response to the sclerotherapy. Different types of sclerosants were used as there is no consensus from previous literature on which is the most appropriate one and sometimes it depends on which fibrosing agent is available at the time of the procedure [19,20]. We didn't notice any differences in the safety or the response with regards to the sclerosants, however, lately we used doxycycline more frequently specially when the size of the cyst is large and it needs more volume to fill the cavity as the other sclerosant like STS3% has limited dose in each session which cannot be exceeded (we do not exceed 10ml pure STS per session). We didn't notice any differences in response with regards to the site involved whether it is in axial or appendicular skeleton, though the majority of our patients had their lesion in the appendicular skeleton.



**Figure 1a:** Complete ossification of the 4<sup>th</sup> metatarsal in patient 1: (A,B) Preoperative X-ray and MRI. (C) Postoperative X-ray



**Figure 1b:** Partial ossification of the sacrum in patient 9: (A,B) Preoperative X-ray and MRI. (C) Postoperative X-ray

### Conclusion

Due to our limited and small sample size, we recommend further studies in other centers, follow-up, as well as further recruitments of patients to ensure the effectiveness of this minimally invasive procedure.

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