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## Case report

# Selective arterial embolization of aneurysmal bone cyst in the pubic bone: A possible primary treatment

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## ABSTRACT

Aneurysmal bone cyst is a benign highly vascular lesion that occurs in children. Traditionally ABCs were treated by surgical resection. However, lesions at difficult to access anatomical locations such as the pelvis have higher morbidity when treated surgically. Recently with the advances in endovascular treatment selective arterial embolization became a promising option for primary treatment of ABC. The authors present a case of a 14-year-old female with a pelvic ABC that was successfully treated by selective arterial embolization. Selective arterial embolization is a cost-efficient way of managing ABC especially in cases where surgical treatment carries high risk.

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## Introduction

An aneurysmal bone cyst (ABC) is a rare benign expansile highly vascular osseous lesion that is locally destructive with a high recurrence rate [1]. It usually occurs in young individuals as either a primary lesion or as a secondary lesion arising from other osseous lesions. ABC can affect any bone; however, it is estimated that 12% of the lesions occur in the pelvis representing half of all flat bone ABCs [2,3]. Curettage and

surgical resection were considered the treatment of choice for ABC; however, lesions in the pelvis are challenging to treat with traditional surgical procedures due to their inaccessible location, large size, and proximity of the lesion to neurovascular structures [4]. Therefore, other less invasive treatment options were introduced, including cryotherapy, intralesional injection, and selective arterial embolization [5–7].

In the past, selective arterial embolization was used as a preprocedural measure to decrease chances of hemorrhage,

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**Fig. 1 – Frontal radiograph showing expansile well defined lytic lesion in the left superior pubic ramus.**

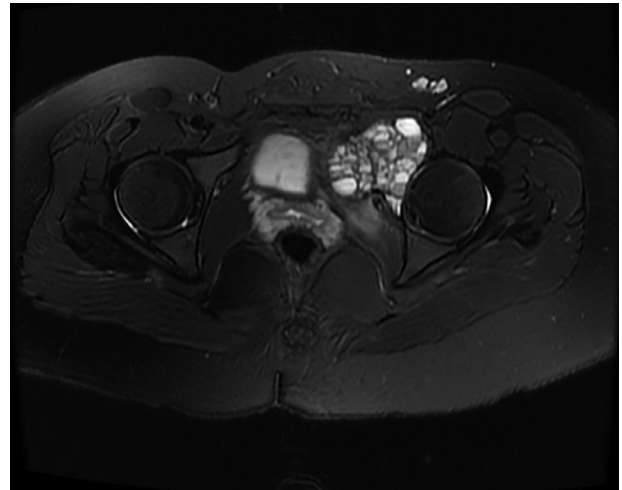
but recently few publications reported using selective arterial embolization as a primary treatment especially at difficult to access anatomical locations [7–11]. Here we present a case of a 14 years old female presenting with a pelvic aneurysmal bone cyst, which was successfully treated by selective arterial embolization as a primary treatment.

### Case Report

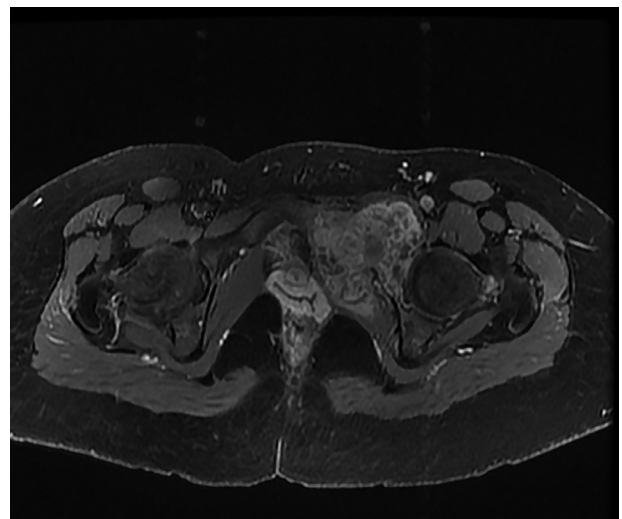
A 14-year-old female patient, previously healthy, presented to the clinic with left hip pain dull in nature for 1 year with no progression and no other constitutional symptoms. Physical examination findings revealed tenderness and swelling over the left lower pelvis, the rest of the examinations were unremarkable. A pelvic radiograph was ordered for the patient and showed an expansile well defined lytic lesion in the left superior pubic ramus (Fig. 1). Further evaluation with pelvic MRI was done, which showed the lesion's multicystic nature with fluid-fluid level and enhancement of the internal septations in post gadolinium images (Figs. 2 and 3). The main considered diagnosis was primary aneurysmal bone cyst; however, other differentials had to be ruled out. Therefore, we did a CT guided biopsy (Fig. 4); histological examination of the biopsy confirmed the radiological diagnosis of a primary aneurysmal bone cyst.

Treatment options and outcomes were discussed with the parents. Due to the age of the patient, anatomical location, and the extensive nature of the lesion, as well as the risk of surgical treatment, selective arterial embolization (SAE) was preferred. The parents' consent was granted for the procedure.

Under fully sterile technique and Xylocaine 1% as local anesthesia, diagnostic digital subtraction angiography (DSA) was performed to identify the feeding vessels and selective embolization through the right common femoral.



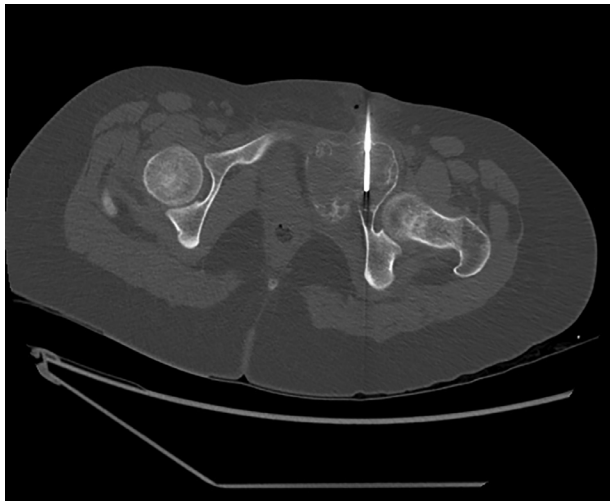
**Fig. 2 – MRI of the same lesion showing its multicystic nature with fluid-fluid level.**



**Fig. 3 – MRI of the same lesion showing the enhancement of the internal septations in post gadolinium images.**

The procedure was initially started using the Seldinger technique, access was granted, and a 5 French sheath was placed. A 5 French catheter and a micro-catheter were used to reach the pathological feeding arteries. The main feeding artery of the left superior rami hypervascular lesion was a Corona Mortis artery arising from the left inferior epigastric artery, which was successfully embolized with 300–500  $\mu$  Polyvinyl alcohol (PVA) particles. After that, identification of the other smaller feeding arteries was made, they originated from the left deep circumflex iliac artery; they were also embolized with 300–500  $\mu$  PVA particles. The final angiogram showed complete devascularization of the lesion (Fig. 5).

The procedure had no immediate complications, the patient tolerated the procedure well, and she was discharged on the same day.



**Fig. 4 – CT guided biopsy of the lesion was done by musculoskeletal radiologist, the tissue sample was sent to pathology which confirmed the diagnosis of ABC.**

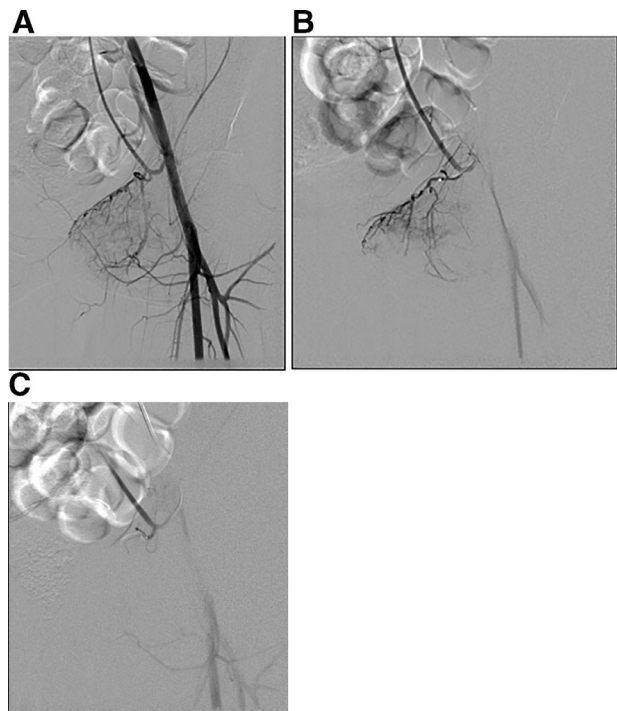
Routine follow up was done in the clinic, and the gradual improvement of the symptoms was observed. Follow-up by radiograph was done every three months initially, then every six months, then once a year, this continued for four years, which showed a gradual reduction in the size of the lesion with progressive trabecular bone formation. Also, 2 postembolization MRIs were done. The first MRI was in 1 year after embolization, and the second was done 3 years after that. The lesion showed a marked decrease in size and reduction of cystic appearance. Therefore, we decided there was no need for another SAE. At 4 years, the patient reported she was symptom-free, radiological findings showed no evidence of local recurrence (Fig. 6).

## Discussion

In this case report, we explore the role of selective arterial embolization as a primary treatment for an aneurysmal bone cyst. Primary ABC is a rare benign, highly vascular lesion that can occur in the pelvis [12,13]. The diagnosis can be made on medical imaging which will show on MRI an expansile lesion with fluid-fluid level [14,15]. However, other differential diagnoses in the pediatric age group should include unicameral bone cyst, osteoblastoma, fibrous dysplasia, nonossifying fibroma, and telangiectatic osteosarcoma [16,17]. Therefore biopsy and histological examination are essential [1,18,19].

Given the highly vascular nature of aneurysmal bone cyst, arterial embolization should be a promising option for the management. Previously selective arterial embolization was initially done to reduce perioperative bleeding; with time and advances in endovascular treatment, the SAE role has been expanded to the definitive treatment. As with our case, the patient was treated by selective arterial embolization as a primary method of treatment.

Aneurysmal bone cysts usually do not have a main supplying artery; instead, they are supplied by more than one abnor-

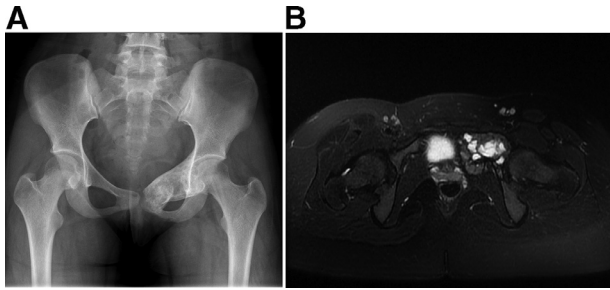


**Fig. 5 – A) Selective left external iliac artery angiogram showing the high vascular lesion, B) Superselection of the main feeding artery angiogram by microcatheter, and C) Post embolization showing complete devascularization of the lesion.**

mal feeding arteries. Evaluation with an angiogram is crucial to identify the feeding arteries, their collaterals, intralesional arteriovenous fistulas, and to prevent the embolization of normal adjacent tissue. Selective arterial embolization is, therefore, essential to only embolize the pathological feeding arteries without affecting the surrounding normal tissue [20–22].

In the literature, there are some publications that showed the positive outcome of SAE, including a reduction in the lesion's size, remineralization, and elimination of pain. For example, in De Cristofaro et al. case series in 1992, which covered 19 patients with ABC treated exclusively with SAE, only 2 cases had a recurrence, and they were treated with a second SAE. The authors suggest using selective arterial embolization as a primary treatment in lesions where surgery risks are high such as in the spine or pelvis [11]. Our patient as well had a large lesion at the left superior rami, and surgery was not preferred due to the high risks. The patient was treated once with no evidence of recurrence in 4 years (Fig. 6).

Rossi G et al. also published a case series of 36 cases of ABC treated by SAE in 2010, 17 of which were in the pelvis. Their results show that 61% of the cases were treated with only one embolization session without recurrence. The authors considered the lesion was healed in 67% of their patients, where the follow-up was more than 2 years [23]. We also did a routine follow-up was for our patient with radiograph and MRI for 4 years, which showed the fast healing that can happen with this age group, and more importantly, it showed that SAE did not cause any bone growth arrest.



**Fig. 6 – A) Frontal radiograph of the pelvis 4 years after treatment showing regression in the size of the lesion with progressive trabecular bone formation and B) MRI of the same lesion 4 years after treatment showing regression in the size and reduction of cystic appearance of the lesion.**

This case report also demonstrates the efficiency and cost to benefit ratio of managing aneurysmal bone cyst with selective arterial embolization. Our patient was admitted for 1 day, did the procedure under local anesthesia, and discharged later the same day. She was able to continue her daily activities and attend school without any reported issues. Therefore, we believe that selective arterial embolization is a safe and successful treatment option for aneurysmal bone cyst, especially in locations where surgery could have significant risks.

#### REFERENCES

- [1] Mankin HJ, Hornicek FJ, Ortiz-Cruz E, Villafuerte J, Gebhardt MC. Aneurysmal bone cyst: a review of 150 patients. *J Clin Oncol* 2005. doi:10.1200/JCO.2005.15.255.
- [2] Erol B, Pill SG, Guttenberg ME, Meyer JS, Dormans JP. Pathologic hip fracture in a 4-year-old boy. *Clin Orthopaed Rel Res* 2002. doi:10.1097/00003086-200210000-00038.
- [3] Vergel De Dios AM, Bond JR, Shives TC, McLeod RA, Unni KK. Aneurysmal bone cyst. A clinicopathologic study of 238 cases. *Cancer* 1992. doi:10.1002/1097-0142(19920615)69:12(2921::AID-CNCR2820691210)3.0.CO;2-E.
- [4] Papagelopoulos PJ, Choudhury SN, Frassica FJ, Bond JR, Unni KK, Sim FH. Treatment of aneurysmal bone cysts of the pelvis and sacrum. *J Bone Jt Surg - Ser A* 2001. doi:10.2106/00004623-200111000-00009.
- [5] Cottalorda J, Bourelle S. Current treatments of primary aneurysmal bone cysts. *J Pediatr Orthop Part B* 2006. doi:10.1097/01.bpb.0000210588.50899.29.
- [6] Boriani S, Lo SFL, Puvanesarajah V. Aneurysmal bone cysts of the spine: Treatment options and considerations. *J Neurooncol* 2014. doi:10.1007/s11060-014-1540-0.
- [7] Doss VT, Weaver J, Didier S, Arthur AS. Serial endovascular embolization as stand-alone treatment of a sacral aneurysmal bone cyst: case report. *J Neurosurg Spine* 2014. doi:10.3171/2013.11.SPINE13412.
- [8] Dick HM, Bigliani LU, Michelsen WJ, Johnston AD, Stinchfield FE. Adjuvant arterial embolization in the treatment of benign primary bone tumors in children. *Clin Orthop Relat Res* 1979. doi:10.1097/00003086-197903000-00023.
- [9] Yıldırım E, Ersözlü S, Kirbaş I, Özgür AF, Akkaya T, Karadeli E. Treatment of pelvic aneurysmal bone cysts in two children: selective arterial embolization as an adjunct to curettage and bone grafting. *Diagnostic Interv Radiol* 2007;13(1):49–52.
- [10] Rossi G, Mavrogenis AF, Rimondi E. Selective arterial embolisation for bone tumours: experience of 454 cases. *Radiol Medica* 2011. doi:10.1007/s11547-011-0670-0.
- [11] De Cristofaro R, Biagini R, Boriani S. Selective arterial embolization in the treatment of aneurysmal bone cyst and angioma of bone. *Skeletal Radiol* 1992. doi:10.1007/BF00195235.
- [12] Cottalorda J, Kohler R, Sales De Gauzy J. Epidemiology of aneurysmal bone cyst in children: A multicenter study and literature review. *J Pediatr Orthop Part B* 2004. doi:10.1097/01202412-200411000-00008.
- [13] Dormans JP, Hanna BG, Johnston DR, Khurana JS. Surgical treatment and recurrence rate of aneurysmal bone cysts in children. *Clin Orthop Relat Res* 2004. doi:10.1097/01.blo.0000126336.46604.e1.
- [14] Keenan S, Bui-Mansfield LT. Musculoskeletal lesions with fluid-fluid level: A pictorial essay. *J Comput Assist Tomogr* 2006. doi:10.1097/00004728-200605000-00029.
- [15] Mahnken AH, Nolte-Ernsting CCA, Wildberger JE. Aneurysmal bone cyst: value of MR imaging and conventional radiography. *Eur Radiol* 2003. doi:10.1007/s00330-002-1668-8.
- [16] Campanacci M. Bone and soft tissue tumors. 2nd ed. P. 639-651; 1990. doi:10.1007/978-3-7091-3846-5\_41
- [17] Zehetgruber H, Bittner B, Gruber D. Prevalence of aneurysmal and solitary bone cysts in young patients. *Clin Orthop Relat Res* 2005. doi:10.1097/01.blo.0000173256.85016.c4.
- [18] Cottalorda J, Bourelle S. Modern concepts of primary aneurysmal bone cyst. *Arch Orthop Trauma Surg* 2007. doi:10.1007/s00402-006-0223-5.
- [19] Campanacci M, Capanna R, Picci P. Unicameral and aneurysmal bone cysts. *Clin Orthop Relat Res* 1986. doi:10.1097/00003086-198603000-00004.
- [20] Kónya A, Szendrői M. Aneurysmal bone cysts treated by superselective embolization. *Skeletal Radiol* 1992. doi:10.1007/BF00242130.
- [21] Mohit AA, Eskridge J, Ellenbogen R, Shaffrey CI. Aneurysmal bone cyst of the atlas: Successful treatment through selective arterial embolization: Case report. *Neurosurgery* 2004. doi:10.1227/01.NEU.0000137279.58768.7E.
- [22] Rossi G, Angelini A, Mavrogenis AF, Rimondi E, Ruggieri P. Successful treatment of aneurysmal bone cyst of the hip in a child by selective transcatheter arterial embolization. *J Vasc Interv Radiol* 2010. doi:10.1016/j.jvir.2010.06.016.
- [23] Rossi G, Rimondi E, Bartalena T. Selective arterial embolization of 36 aneurysmal bone cysts of the skeleton with N-2-butyl cyanoacrylate. *Skeletal Radiol* 2010. doi:10.1007/s00256-009-0757-z.