

Selective Arterial Embolization (SAE) in Aneurysmal Bone Cyst (ABC)

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Abstract

Aim: The aim of the study was to evaluate the success of selective arterial embolization (SAE) treatment in aneurysmal bone cysts (ABC).

Materials and Methods: Patients were referred to our clinic between January 2013 and December 2019 with a pathological diagnosis of ABC were analyzed retrospectively. The study was completed with 18 patients (9 male, 9 female) (7-59 years). Six ABCs were located in the axial skeleton and twelve were in the appendicular skeleton. SAE was done with particulate or liquid embolic agents with a coaxial catheter system. Demographic data of the patients, and the correlation between lesion size and total embolization were analyzed. Complications related to the angiographic procedure were also examined. Patients were considered as complete responders if no recurrence was noticed at two-year follow-up after SAE. Patients with shorter follow-up without recurrence were considered asymptomatic; $p < 0.05$ was considered as statistically significant.

Results: In this study population, a single embolization was performed in ten patients (55.56%), two consecutive embolizations in six patients (33.33%), and three consecutive embolizations in two patients (11.11%). A low correlation was observed between embolization numbers and lesion sizes ($k = 0.35$). The mean follow-up period of the patients who underwent SAE was 33.76 months (6-61 months). The treatment response was achieved in 88.9% (n: 16) of the patients who received SAE. Surgery was performed in two patients (11.1%) due to recurrence. There were no major complications recorded such as skin necrosis or paresthesia.

Conclusion: Due to its low complication rates, easy reproducibility, and similar success rate with surgery, SAE is a reliable method for the treatment of ABC.

Keywords: Aneurysmal bone cyst; embolization; selective arterial embolization

INTRODUCTION

Aneurysmal bone cyst (ABC) was first defined by Lichtenstein in 1942 and constitutes 1% of all primary bone tumors in pathological specimens (1-3). ABC is a benign bone tumor and is a lytic lesion containing cystic components in radiological examinations (4-6). Lesions often present with pain and swelling for less than three months (5,7). Its etiology is not fully understood, and it has been hypothesized that ABC may develop secondary to trauma or primary benign and malignant tumors of the bone (1,8). Most ABCs (95%) occur in the first three decades and are more common in women than in men (1,7,8). Percutaneous interventions, surgical procedures, and selective arterial embolization (SAE) can be used in treatment (8, 9-13). Radiotherapy is rare due to complications that may be encountered in the late period. (14). SAE is the preferred option in ABC treatment because it is reproducible and has a high success rate. Still, the

success rates of SAE decrease in patients under the age of 15, female patients, and patients with lesions larger than 5 cm (7,14).

The aim of this study is to evaluate the radiological and clinical success rates after SAE was applied in ABC and to understand the role of SAE in ABC treatment.

MATERIALS and METHODS

This retrospective study was conducted with 21 patients (10 men, 11 women) diagnosed with ABC according to imaging findings between January 2013 and December 2019 after receiving approval from the internal review board (I6-383-21). Informed consent was obtained from all patients or their legal guardians included in the study.

Inclusion criteria were having a pathological diagnosis of ABC and showing hypervascularity in angiographic examination. ABCs without vascularity in angiographic examination and patients diagnosed as ABC with clinical

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and radiological findings without pathologic correlation were excluded from study.

All embolization procedures were provided with a coaxial catheter system to achieve super-selective catheterization. Patients were embolized with spherical microspheres, N-butyl cyanoacrylate (NBCA), or Onyx. Age, gender, lesion size, lesion localization, total embolizations, changes in the radiological appearance of the lesions, and follow-up periods after treatment were recorded and analyzed. Correlation between these data was investigated. Treatment success was evaluated according to clinical and radiological response. Improvement in pain was considered to be a clinical success. Reduction in tumor size by more than 25% or ossification of more than 25% of the tumor was considered radiological success. Patients who underwent surgery after SAE were considered to have recurred.

Angiography procedure and SAE

All embolization procedures were performed with an Artis Zee (Siemens Healthcare, Erlangen, Germany) angiography device. Right or left femoral artery catheterization with a 5 F vascular sheath were preferred for all patients. Angiography procedures were performed after local anesthesia was applied to the femoral region, and general anesthesia was preferred in pediatric patients. The contralateral approach was used to catheterize the feeders of ABCs located in the pelvic region and lower extremities. After selective catheterization of the intended artery, superselective catheterization was provided with a combination of microcatheters (2.4 Fr, 2.7 Fr) and micro-guidewires (0.014 ", 0.016 ", 0.21 "). A non-ionic contrast material (350 mg / mL) was used during angiographic examinations. Particulate (300-500 μ m) or liquid embolics (NBCA, Onyx) were used separately or in combination with the embolization procedures; 25% dilution was preferred for NBCA applications. NBCA was applied using the "sandwich method" together with 5% dextrose solution (14). Hemostasis of the femoral artery after the removal of the sheath was achieved by manual compression. Patients' follow-up was performed according to our institutional protocol and as follows: clinical examination and radiological imaging quarterly in the first year, semiannually for the second year, and once annually for the following years. Recurrence of pain symptoms or increased lesion size in follow-up after SAE were considered as a recurrence, and no recurrence after 2 years of follow-up was considered to be a complete recovery. Patients with a shorter follow-up period were considered asymptomatic. The Society of Interventional Radiology guidelines were used in the evaluation of angiography-related complications (15,16).

Statistical analysis

Descriptive data on age were shown as an average, and categorical data (number of cases) were shown as a percentage (%). Age distributions were compared across different gender groups via Student's t-test. The mean, median, minimum and maximum values were used

for descriptive statistics. Numbers and percentages were used for discrete data. The relationship between categorical variables was analyzed using Pearson's Chi-square test. The conformity of the data to the normal distribution was tested for comparison of continuous data in patients undergoing SAE. Student's t-test was used for patients with normal distribution, and the Mann Whitney U test was used for patients who did not. IBM SPSS (IBM Corp. Released 2011. IBM SPSS Statistics for Windows, Version 20.0. Armonk, NY: IBM Corp.) was used to evaluate the statistical data. Here, $p < 0.05$ was considered statistically significant.

RESULTS

This study was conducted with 18 patients (nine men and nine women). Three patients were not included to the study due to lack of pathologic diagnosis or a lack of vascularity at angiographic imaging. The mean age of the patients was 23.7 years (7-59 years). Six lesions were located in the axial skeleton and twelve lesions were in the appendicular skeleton. The mean lesion size was calculated as 134 cm³ (2.5-11 cm). Eleven lesions were smaller than 5 cm, and seven lesions were larger than 5 cm in maximal diameter (Table 1).

Table 1. Demographic distribution of the patients

Patients (n)	18 (9:male, 9:female)	
Age (mean)	23.7 years (7-59 years)	
Mean lesion size (cm ³)	134 (2.5-11cm)	
Lesions > 5 cm	7	
Lesions < 5 cm	11	
Lesion localization	axial skeleton	appendicular skeleton
	6	12

A total of 28 embolization procedures were applied to the patients; where, 26 embolization procedures were performed with spherical microspheres, one embolization procedure with spherical microspheres + NBCA, and one embolization procedure with Onyx.

A single embolization procedure was performed in 10 patients (55.6%), two sequential embolization procedures (33.3%) in six patients, and three consecutive embolization procedures in two patients (11.1%). There was no correlation between the gender of the patients and the number of embolizations ($k=0$). A low correlation was found between lesion sizes and embolization numbers ($k=0.35$).

Surgical treatment was performed after SAE due to the persistence of pain symptoms in two female patients (11.1%). No recurrence was observed in the follow-up of these patients (2-5 years).

The mean follow-up period of the patients included in the study was 33.76 months (6-61 months). Follow-up periods were similar for both gender ($p=0.739$).

During follow-up, complete recovery (66.7%) was obtained in seven males and five females (Figure 1). Four patients had regression in symptoms during their 6-18 months of follow-up and were considered asymptomatic (22.2%) (Figure 2).

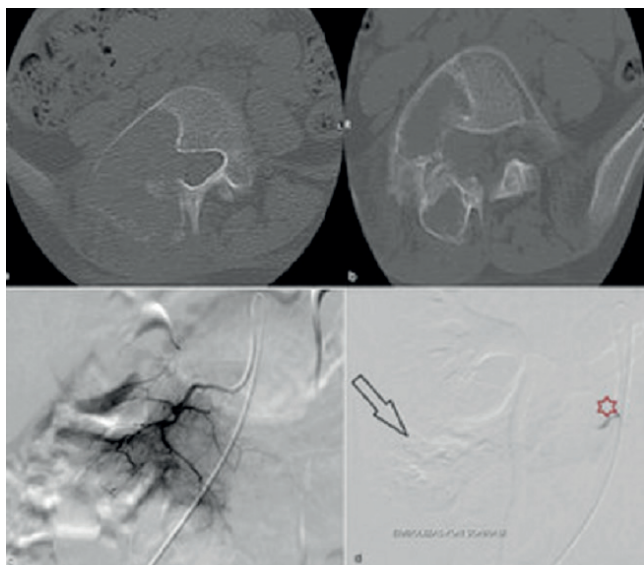


Figure 1. Complete clinical response with successful radiological response in a female patient with an expansile ABC localized in the right half of the L5 vertebra (images obtained during 60th month follow-up). a; Computed tomography images obtained before embolization, b; Control images obtained 60 months after successful embolization, c; DSA images of hypervascular ABC obtained before embolization, d; Control angiography images after embolization with spherical microspheres



Figure 2. ABC located in the sacral vertebra; MR images obtained in 18 months follow-up. a; Pre-operative computed tomography image, b; Image after embolization with spherical microsphere and NBCA, c; MR images obtained at the 18th month (reduction in lesion size)

No complications such as paresthesia or skin necrosis were observed in any of the patients. A small hematoma (<3 cm) developed in two patients, and one patient had a pseudoaneurysm; all of these were at the femoral puncture site. Post-embolization syndrome was seen in four patients.

DISCUSSION

SAE is a valuable treatment method that has been shown to be effective in ABC. The most important factors that reduce the success of SAE treatment are lesions with a maximum diameter greater than 5 cm, female gender, and being younger than 15 years old (7, 14). These results are compatible with the literature.

Approximately 10% of ABCs did not demonstrate tumoral vascularization in angiographic imaging (14). Here, two patients were referred to our clinic with an ABC diagnosis according to pathology and did not demonstrate tumor vascularization.

It has been reported that 90% of the recurrences that develop after ABC treatment develop in the first two years of follow-up (12). Follow-up for at least two years will be required for full recovery after SAE of ABC (7). We achieve complete recovery in 12 patients (66.7%) during follow-up regardless of the number of embolizations. Despite the reduction in lesion size, partial ossification, and improvements in clinical symptoms, four patients (22.2%) were considered asymptomatic because the follow-up period was shorter than 2 years. No recurrence was observed in the follow-up of two patients (11.1%) who underwent curettage and bone grafting after embolization.

Rossi et al. studied 102 patients in 2017 and reported that 81.8% of patients achieved clinical and radiological response after SAE with NBCA regardless of the follow-up period. The rate of surgery after embolization in the same study was calculated as 18.2% (n=16) (14). The response rate was calculated as 88.9% in our study independent of the follow-up period. The surgery rate was 11.1% (n=2) in this study. These findings show that the results of our study are similar to the literature.

Standard ABC treatment is surgery, and the postoperative recurrence rates were recorded between 10-71% in the literature (12, 17, 18). Percutaneous interventions are alternative options in the treatment of ABC. Zarzour et al. reported no recurrence at annual follow-up of 20 ABC patients treated with percutaneous radiofrequency ablation in 2018. (19). Considering that recurrences frequently occur in the first two years after treatment in ABCs, the reliability of this study is insufficient in terms of recurrence rates. In contrast, Shiels et al. reported that the recurrence rates after more than two years of follow-up after 'off-label doxycycline treatment' in ABC were 5% in 2013 (18). The recurrence rates in these studies were lower than our results.

It has been reported that sclerotherapy with polidocanol can provide successful results including percutaneous treatment options in the treatment of ABC. Rastogi et al. studied 72 patients in 2006, and the radiological healing rate was 76.6% after 1-5 sessions of treatment with polidocanol in the follow-up. The recurrence rates were recorded as 2.6% after an average of 11.8 months of treatment (12). Despite the short follow-up period, these findings are similar to our study.

When the relationship between lesion size, age, and recurrence rate were evaluated, the recurrence rates after treatment were lower for ABCs smaller than 5 cm in maximum diameter, male patients, and subjects older than 15 years (7, 14). Although the number of patients included in our study is insufficient to show a statistical difference, our results are similar to the literature.

The rate of permanent complications after SAE in the literature is as high as 4.5% (14). Six patients had category A or B complications, and one patient had a category C complication according to SIR guidelines (15). The patient who presented with a pseudoaneurysm at the femoral puncture site was treated with compression of the pseudoaneurysm under ultrasound guidance because the size of the pseudoaneurysm did not decrease in weekly follow-up. The hematoma resolved after a few weeks of surveillance. Four patients who developed post-embolization syndrome recovered in the following days. None of the patients in our study developed complications that would cause permanent sequelae. We believe that avoiding excessively aggressive embolization and selecting the correct particle size is key to preventing permanent sequelae while treating ABC with SAE. Different embolic agents used during SAE may also cause complication rate differences.

The most important limitation of our study is that it is retrospective with a limited patient size. These limitations reduced our studies' statistical power. Another limitation is the lack of a comparison group treated with a different treatment method. Studies that investigate the effect of combination treatments with SAE in ABC may be beneficial.

CONCLUSION

SAE is a reproducible and reliable treatment option that can be preferred to surgery in the treatment of ABC.

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REFERENCES

- Freiberg AA, Loder RT, Heidelberger KP, et al. Aneurysmal bone cysts in young children. *J Pediatr Orthop* 1994;14:86-91.
- Jaffe HI, Lichtenstein L. Solitary Unicameral Bone Cyst: With Emphasis On The Roentgen Picture, The Pathologic Appearance And The Pathogenesis. *Arch Surg* 1942;44:1004-25.
- Henrichs MP, Beck L, Gosheger G, et al. Selective arterial Embolisation of Aneurysmal Bone Cysts of the Sacrum: a promising Alternative to Surgery. *Rofo* 2016;188:53-9.
- Marcove RC, Sheth DS, Takemoto S, et al. The treatment of aneurysmal bone cyst. *Clin Orthop Relat Res* 1995;311:157-63.
- Boriani S, De Iure F, Campanacci L, et al. Aneurysmal bone cyst of the mobile spine: report on 41 cases. *Spine* 2001;26:27-35.
- Rapp TB, Ward JP, Alaia MJ. Aneurysmal bone cyst. *J Am Acad Orthop Surg* 2012;20:233-41.
- Rossi G, Rimondi E, Bartalena T, et al. Selective arterial embolization of 36 aneurysmal bone cysts of the skeleton with N-2-butyl cyanoacrylate. *Skeletal Radiol* 2010;39:161-7.
- Bonakdarpour A, Levy WM, Aegerter E. Primary and secondary aneurysmal bone cyst: a radiological study of 75 cases. *Radiology* 1978;126:75-83.
- Campanacci M, Capanna R, Picci P. Unicameral and aneurysmal bone cysts. *Clin Orthop Relat Res* 1986;204:25-36.
- Meyer S, Reinhard H, Graf N, et al. Arterial embolization of a secondary aneurysmatic bone cyst of the thoracic spine prior to surgical excision in a 15-year-old girl. *Eur J Radiol* 2002;43:79-81.
- Falappa P, Fassari FM, Fanelli A, et al. Aneurysmal bone cysts: treatment with direct percutaneous Ethibloc injection: long-term results. *Cardiovasc Intervent Radiol* 2002;25:282-90.
- Rastogi S, Varshney MK, Trikha V, et al. Treatment of aneurysmal bone cysts with percutaneous sclerotherapy using polidocanol. A review of 72 cases with long-term follow-up. *J Bone Joint Surg Br* 2006;88:1212-6.
- Rossi G, Mavrogenis AF, Rimondi E, et al. Selective arterial embolisation for bone tumours: experience of 454 cases. *Radiol Med* 2011;116:793-808.
- Rossi G, Mavrogenis AF, Facchini G, et al. How effective is embolization with N-2-butyl-cyanoacrylate for aneurysmal bone cysts? *Int Orthop* 2017;41:1685-92.
- Khalilzadeh O, Baerlocher MO, Shyn PB, et al. Proposal of a new adverse event classification by the Society of Interventional Radiology Standards of Practice Committee. *J Vasc Interv Radiol* 2017;28:1432-7.
- Angle JF, Siddiqi NH, Wallace MJ, et al; Society of Interventional Radiology Standards of Practice Committee. Quality improvement guidelines for percutaneous transcatheter embolization: Society of Interventional Radiology Standards of Practice Committee. *J Vasc Interv Radiol* 2010;21:1479-86
- Biesecker JL, Marcove RC, Huvos AG, et al. Aneurysmal bone cysts. A clinicopathologic study of 66 cases. *Cancer* 1970;26:615-25.
- Shiels WE, Mayerson JL. Percutaneous doxycycline treatment of aneurysmal bone cysts with low recurrence rate: a preliminary report. *Clin Orthop Relat Res* 2013;471:2675-83.
- Zarzour OA, Santiago FR, Serrano NO, et al. CT-guided radiofrequency ablation in patients with aneurysmal bone cysts. *Eur J Radiol* 2018;100:116-23.