

Recurrence Rates in Twenty Four Patients with Pelvic Aneurysmal Bone Cyst with a Minimum Follow-up of Two Years

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Abstract

Background: Pelvic Aneurysmal Bone Cysts (ABC) is challenging lesions for the orthopedic surgeon. Compared to ABCs located in the extremities, pelvic ABCs present unique problems due to the difficulty of exposures or proximity to critical neurovascular structures. Common treatment options include intralesional surgery (curettage with or without cementing, phenol, auto or allografting), resection, arterial embolization in select cases. The aim of this study is to present our results regarding recurrence rates in a cohort of 24 patients.

Case presentation: Twenty-four patients operated for pelvic ABC from 1992 to 2018 were retrospectively reviewed. Presenting symptoms, application of preoperative embolization, type of operation, detection of recurrence, time to recurrence and further management and follow-up times were noted. Average follow-up was 66 months (24-200).

Results: Lesions were localized in the periacetabular region in 3 cases (12.5%), pubic rami in 5 cases (20.8%), iliac wing in 9 cases (37.5%) and sacrum in 7 cases (29.2%). Preoperative angioembolization was used in 7 cases (29.2%). Recurrence was observed in 5 (20.8%) of 24 cases. Recurrence occurred at an average of 27.7 months (6-84, median 18.4) after the primary surgery. Chi-square test for using of bone cement and rate of recurrence was not statistically significant ($p > 0.05$).

Conclusion: Through careful planning, comprehensive imaging and meticulous surgical treatment predictably low recurrence rates can be achieved.

Keywords: Aneurysmal bone cyst; Resection; Extended curettage; Cementing

Introduction

Aneurysmal bone cysts (ABC), first described by Lichtenstein in 1942 [1], are rare tumors that expand the bone and may cause pain, swelling and pathological fractures. Its incidence is reported to be ranging from 1.4 to 3.2 per 1,000,000 individuals [2,3]. Some authors report it is more common in males [2,3], while there are also studies that report a female predilection [4,5]. A 1,191 patient case series from the Rizzoli Case Archive, on the other hand, concluded that there is no gender predilection [6]. They are most commonly encountered in the second decade and are rare after the sixth decade. ABCs can be primary lesions in about 70% of cases and secondary in the remaining 30% [7]. They can be seen in any bone but are most commonly found in the metaphysis of long bones. Common sites include proximal humerus, proximal femur, proximal tibia, pelvis. The Enneking staging system, which is based on radiographic characteristics of the tumor-host margin is typically used for radiological classification [8]. Stage 1 lesions have well-demarcated borders and are considered

latent whereas Stage 2 and Stage 3 lesions have indistinct borders and are respectively considered active and aggressive. Common treatment options include intralesional surgery (curettage ± cementing, phenol, auto or allografting), resection or arterial embolization. Pelvic ABCs can be large at presentation and highly vascular, which requires extensive preoperative planning and in some cases use of adjuvant angioembolization [9]. Other less common treatment options include curettage, bone marrow injection, sclerotherapy, cryotherapy and radiotherapy. Local recurrence rate in the literature is reported to be about 20% [5,10-13]. ABCs can be especially problematic when in the axial skeleton, due to the difficulty of exposures or proximity to critical neurovascular structures. We present our results regarding pelvic ABCs operated over more than two decades with regards to different treatment techniques and recurrence rates.

Case Report

IRB approval was obtained for this study. Twenty-four patients operated for pelvic ABC from 1992 to 2018 were retrospectively reviewed. 7 patients were referred to our institution after the biopsy result suggested ABC. In these cases, all biopsies were reevaluated by

an experienced musculoskeletal pathologist. Surgical planning was done with comprehensive radiological examination of the pelvis in all cases (X-ray, CT as well as MRI for all cases from 1995 onwards). All radiographic studies were evaluated jointly with a musculoskeletal radiologist. The lesions were staged according to Enneking staging system [8]. Procedures were grouped as curettage and bone grafting, extended curettage, and resection. Extended curettage consisted of high speed burr, phenol and thermal ablation with electrocautery. Bone cement was used in a total of 8 cases (Figure 1). 1 case with bone grafting and 7 cases with extended curettage. 14 of the patients were male (58.3%) and 10 were female (41.7%). Average age at the time of index operation was 24.6 (9-56). All patients presented with pain while swelling was present in only 7 patients (29%). Application of preoperative embolization, type of operation, detection of recurrence, time to recurrence and further management and follow-up times were noted. Average follow-up was 66 months (24-200).

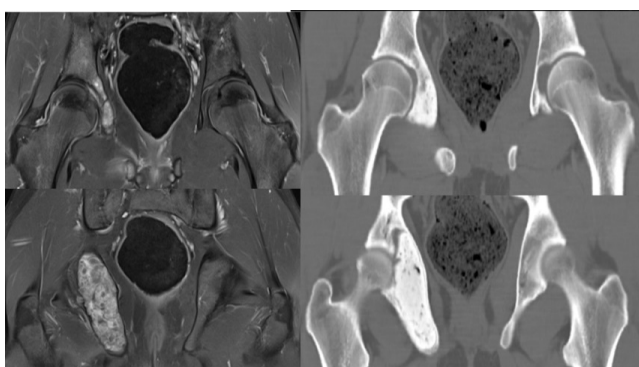


Figure 1: AMS interventions by clinical pharmacists from Jan 2018-Dec 2019.

Results

The demographics, symptomatology and radiological data are presented in Table 1. Lesions were localized in the periacetabular region in 3 cases (12.5%), pubic rami in 5 cases (20.8%), iliac wing in 9 cases (37.5%) and sacrum in 7 cases (29.2%). Preoperative angioembolization was used in 7 cases (29.2%).

ID	Sex	Age	Symptoms	Location	MSTS grade	Trabeculations (XR)	Fluid levels (MRI)
1	F	26	P	Ischion	3	+	+
2	M	22	P	Pubic ramus	3	+	-
3	M	11	P+S	Iliac wing	3	+	+
4	F	21	P+S	Iliac wing	3	-	+
5	F	25	P	Sacrum	2	-	-
6	M	14	P	Iliac wing	2	-	-
7	M	33	P	Sacrum	3	-	-
8	M	56	P	Periacetabular region	2	-	-

9	F	16	P+S	Pubic ramus	3	+	-
10	F	18	P	Pubic ramus	2	+	-
11	M	24	P	Sacrum	3	-	-
12	F	26	P	Iliac wing	2	-	-
13	F	28	S	Iliac wing	2	+	-
14	F	44	P	Pubic ramus	3	-	-
15	F	9	P+S	Iliac wing	3	+	+
16	M	25	P+S	Iliac wing	2	+	+
17	F	34	P+S	Sacrum	3	+	-
18	M	18	P	Periacetabular region	2	-	-
19	M	25	P	Iliac wing	2	-	-
20	M	27	P	Iliac wing	3	-	+
21	M	24	P	Sacrum	3	-	-
22	M	32	P	Sacrum	3	-	-
23	M	11	P	Pubic ramus	2	-	-
24	M	18	P	Sacrum	2	-	-

Note: P=pain, L=limp, S=swelling

Table 1: Demographic, presentation and radiologic data.

17 patients had Enneking Stage 2 lesions whereas 7 patients had stage-3 lesions. Patients were treated with curettage+bone grafting in 3 cases (12.5%) (Figure 2), resection in 7 cases (29.2%) (Figure 3). Recurrence was observed in 5 (20.8%) of 24 cases. Recurrence occurred at an average of 27.7 months (6-84, median 18.4) after the primary surgery. Of the 5 cases with recurrence, 2 were treated with curettage and bone grafting without bone cement, 2 with extended curettage without bone cement and 1 with extended curettage and bone cement. Chi-square test comparing usage of bone cement versus recurrence was not statistically significant ($p>0.05$).

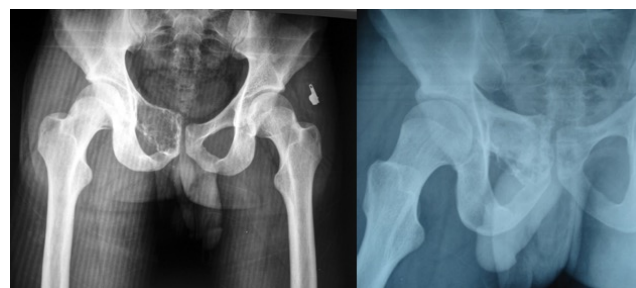


Figure 2: AMS interventions by clinical pharmacists from Jan 2018-Dec 2019.

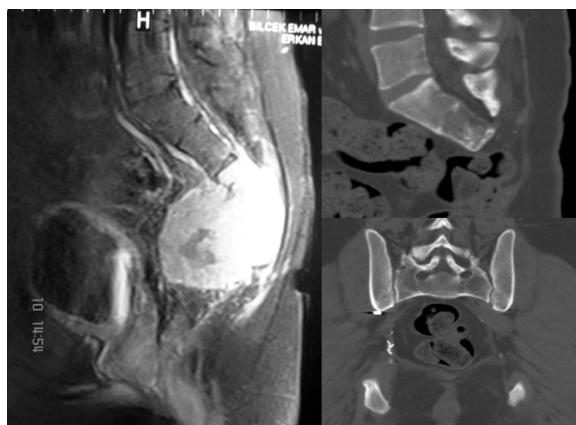


Figure 3: AMS interventions by clinical pharmacists from Jan 2018-Dec 2019.

Two patients with recurrence denied further intervention. One of them was still under follow-up with stable disease at the time this study was prepared while the other was lost to follow-up 10 years after the index operation. Two patients were treated with selective arterial embolization with no further recurrence. One patient was operated with curettage and allografting and had a second local recurrence four months after the second operation. Patient was re-operated extended curettage and bone cement and have since been disease free.

Discussion

Presentation of pelvic ABC is usually with pain and/or limping [14]. Similar to our series, Papagelopoulos et al. reported that 95% of the patients presented with pain as the main symptom. Regarding the location within the pelvis, our results are also in accordance with the literature. In the same study by Papagelopoulos, out of 40 patients, 7 were located at pubic rami (17.5%), 16 were periacetabular (40%), 5 were located in the iliac wing (12.5%) and 12 were located in the sacrum (30%) [15].

Pelvic ABC's present unique challenges to the orthopedic surgeon for several reasons. Since lesions are commonly encountered in the first two decades, any effort must be made to preserve the growth plates and native joints of the patients while making sure the lesion is thoroughly curetted/resected. The complex anatomy of the pelvis presents unique problems in surgical technique and reconstruction in this regard.

Surgery of lesions that involve the acetabulum and upper sacral vertebrae especially may result in significant morbidity. Although the optimal treatment method is still debated, open curettage with or without bone grafting is a widely accepted mode of treatment [16].

Extended curettage is a combination of mechanical, chemical and thermal means applied to the lesion surface after the lesion is thoroughly curetted [17]. For mechanical purposes high speed burr is widely used. Additional of high speed burr may have been reported to decrease recurrence rates to 15% [18]. For chemical purposes, different adjuvants such as phenol, 96% ethanol and cement can be used. In our case we always prefer phenol. For thermal purposes we use the 'spray' mode of the electrocautery.

Following curettage, both bone grafting and cementing are viable options. We prefer cementing aneurysmal bone cysts whenever possible in our clinical practice since it provides both mechanical and structural support along with thermal ablation. Since ABCs are benign aggressive lesions, bone grafting may be used in children as well. However, even in pediatric cases, unless serious complications regarding limb length discrepancy is anticipated, we consider cementing preferable. Thermal ablative property of the cement in preventing recurrence may be protective from revision surgeries and irreversible functional losses; and thus may justify its use. In pelvic ABC's we are satisfied with our results in cases cemented following extended curettage. In the periacetabular region sometimes the lesion is located almost adjacent into the subchondral bone of the hip joint. In such cases, cement can be used to prevent subchondral bone from collapsing. In such cases, we first protect the subchondral surface of the cartilage from inside by covering it with an absorbable hemostat such as SURGICEL® before cementing. As the cement hardens, we apply slight pressure to the lower extremity towards the hip joint so that the femoral head acts as a mold to the membrane-like subchondral bone. In sacrum, however, the neural roots are in danger from the thermal effects of the cement. Therefore in sacrum after careful dissection of neurovascular structures extended curettage is performed without using cement.

Selective serial angioembolization is also suggested as a method for treating ABCs. It has the advantage of potentially avoiding surgery and its complications while it is reported to have a recurrence rate of 39%-44% [19-21]. We reserve it for inaccessible lesions or lesions with unacceptable surgical morbidity and use it as an adjuvant therapy prior to surgical intervention.

A 2019 review of pelvic and appendicular ABCs concluded that with contemporary treatment a cure rate of 70%-90% is to be expected [22]. Capanna et al. Have reported a recurrence rate of 13% in their series of 23 patients with pelvic ABC [23]. Papagelopoulos et al. Reported a local recurrence rate of 14% in their series pelvic and sacral ABCs [15]. Our recurrence rate is similar (20%). Of the 5 recurrences, 4 occurred within 2 years with a median time of 18,4 months. While the literature also suggests that recurrence occurs mostly within two years [15,23-25], it should be kept in mind that very late recurrences such as in our case may occur, as in our case one case recurred after 7 years.

Conclusion

Pelvic aneurysmal bone cysts can be challenging lesions for the orthopedic surgeon. Through careful planning, comprehensive imaging and meticulous surgical treatment predictably low recurrence rates can be achieved.

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Conflict of Interest

The authors declare that there is no conflict of interest.

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