www.journalmsr.com

الجمعية السعودية لجراحة العظام
Saudi Orthopedic Association

Journal of Musculoskeletal Surgery and Research

Systematic Review Article

Aneurysmal bone cyst of the pelvis: systematic literature review

Ayesha Saeed, MD. FCPS.[1], Elke Rometsch, MSc.[2], Anahi Hurtado Chong, MD. PhD.[2]

[1]Department of Pediatric Orthopedics, The Children's Hospital and The Institute of Child Health, Faisalabad, Pakistan, [2]AO Investigation and Translation Centre, Dubendorf, Switzerland.

***Corresponding author:**
Ayesha Saeed,
Department of Pediatric Orthopedics, The Children's Hospital and The Institute of Child Health, Faisalabad, Pakistan.

drayeshasaeed@gmail.com

Received: 04 November 2022
Accepted: 30 December 2022
EPub Ahead of Print: 27 January 2023
Published: 07 February 2023

DOI
10.25259/JMSR_135_2022

Quick Response Code:

ABSTRACT

Aneurysmal bone cysts (ABCs) are rare benign, vascular, and osteolytic bone lesions. Pelvic ABCs account for 8–12% of these tumors and no clear guidelines for their treatment are available. To the best of our knowledge, this is the first systematic literature review regarding pelvic ABCs. Our objective was to identify treatment modalities and assess bone healing, measured as the degree of radiological ossification. Searches were conducted in PubMed, Cochrane Library, and Web of Science. Based on the scarcity of reports, inclusion criteria were kept broad and included primary or recurrent pelvic ABCs, with a minimum follow-up of 1 year and available information on radiological ossification. Data were extracted at the individual patient level and grouped according to treatment modality. Forty-nine studies reporting on 194 patients were included from the study. The level of evidence was low (29 case reports and 20 retrospective case series), and the reporting of outcomes was inconsistent. Five major treatment groups were identified and divided into 11 subgroups. The largest subgroup was curettage (23%), followed by selective arterial embolization (20%). Most ABCs were located in the ilium. Variations in mean tumor size (4.5–22.2 cm) and degree of ossification (60–100%) depended on the treatment modality. Overall, in 77% of the cases, the cyst ossified completely. Recurrence was reported in 22 patients (11%) and two patients (1%) died during the course of the treatment. This systematic review provides the first comprehensive overview of pelvic ABC treatment modalities and their radiological and clinical outcomes. Neoadjuvant scleroembolic treatments appear to be used most in recent years, but further comparative studies and better quality of reporting are needed to determine their effectiveness.

Keywords: Aneurysmal bone cyst, Benign, Pelvis, Tumor, Selective arterial embolization

INTRODUCTION

Aneurysmal bone cysts (ABCs), also known as "Jaffe-Lichtenstein disease,"[1-3] are rare benign vascular lytic lesions of the bone of unknown etiology.[4,5] The annual incidence of ABC in all bony locations has been reported as 0.14/100,000 people, with slight female preponderance.[6] However, the true incidence is difficult to calculate as some of the ABC cases may have spontaneous regression, and some cases can be clinically silent.[6] Eighty percentages of ABCs occur within the first and second decades of life.[1,7] The clinical course is sometimes unpredictable, and local recurrences are relatively often,[8] with reported recurrence rates from 5% to 40%.[1,9,10]

How to cite this article: Saeed A, Rometsch E, Chong AH. Aneurysmal bone cyst of the pelvis: Systematic literature review. J Musculoskelet Surg Res 2023;7:11-23.

A study describing the anatomical distribution of 897 ABCs found pelvic lesions in 11.6% of the cases, the fourth most frequent site in descending order after the tibia, femur, and vertebra.[11] In spite of accounting for nearly half of all flat bone lesions,[12] pelvic ABCs are extremely rare. Management of pelvic ABC requires careful consideration of special factors, such as the risk of bleeding, proximity to neurovascular structures, relative inaccessibility, and damage to the integrity of weight-bearing structures such as the acetabulum or sacrum.[8-10]

Several treatment modalities have been reported in the scarce literature available concerning pelvic ABCs.[13] Therefore, we performed a systematic literature review with the primary aim of analyzing the main treatments used for this particular disease and their radiological ossification outcomes. The secondary aims were to describe the demographics, tumor location, clinical and functional outcomes, complications, and recurrence rate.

MATERIALS AND METHODS

A systematic literature review on pelvic ABC was conducted on the September 28, 2017, and reported following the Preferred Reporting Items for Systematic Reviews and Meta-Analyses statement.[14,15]

Electronic searches were conducted in PubMed, the Cochrane Library, and the Web of Science. The search strategy used a combination of the following terms: ([Pelvis OR pelvic] AND [ABC [MeSH Terms] OR "aneurysmal bone cyst"]). Additional articles were identified by hand, searching the reference lists of relevant articles such as large case series or literature reviews. A first survey of the identified literature revealed that it was limited to retrospective case series and case reports that provided information at the individual patient level in most cases. The eligibility criteria used for this systematic review were as follows:

Inclusion criteria

The following criteria were included in the study:
- Primary or recurrent ABC located in the pelvis
- Any age
- Any treatment method (including observation)
- A minimum follow-up of 1 year
- Radiological, clinical, and functional outcomes provided by treatment method
- Mention of ossification status
- English language.

Exclusion criteria

The following criteria were excluded from the study:
- Secondary pelvic ABCs
- Information on treatment or outcome for pelvic tumors was not specified.

No date limits were applied. Screening and eligibility assessment was performed by the first author. Neither prospective nor comparative studies met the eligibility criteria. Due to the study designs and level of evidence found, data either on individual patients or grouped by treatment were extracted. The articles' titles, publication year, authors, and number of patients with pelvic ABC were recorded. Articles describing ABCs in multiple locations but containing pelvic zone cases within their study population were also included in the study; only relevant information related to pelvic ABC was used.

Whenever possible, individual patient information on gender, age, location of ABC according to Enneking and Durham anatomic pelvic zones,[16] primary or recurrent ABC, tumor size, Enneking staging,[17] type of treatment, and years of follow-up was recorded.

Rossi *et al.* described the age groups into four categories, so it was excluded from the calculation of the mean age.[18] According to Enneking and Durham,[16] the pelvic zones used to describe tumor location were: 1 = ilium, 2 = acetabulum, 3 = ischium-pubis, and 4 = sacrum. If a tumor extended over more than one zone, the affected zones were combined into a separate category. Given the different ways of measuring and reporting tumor sizes used in the literature, the mean size was calculated using the maximum dimension reported.

The treatment and the outcomes for pelvic ABCs reported in the studies presented a high degree of heterogeneity. After reviewing the extracted data, treatment modalities were categorized into the following groups and subgroups for the qualitative synthesis of results:

1. Observation
2. Adjuvant treatment
    a. Selective arterial embolization (SAE)
    b. Sclerotherapy
    c. Radiotherapy
    d. SAE + Sclerotherapy
3. Minor surgery
    a. Curettage
    b. Extended curettage
4. Major surgery
    a. Marginal excision
    b. Wide excision
5. Combined
    a. Adjuvant + minor surgery
    b. Adjuvant + major surgery

The primary outcome was radiological ossification, defined as either "no evidence of disease" or "complete consolidation of the lesion" with or without exogenous material. "Partial ossification" was considered when terms such as "cystic

residues," "partial ossification between 25% and 75%," or "healed with small and large residual geodes" were used. "No ossification" was considered for terms like "little ossification, defined as ossification of <25%,"[18] or "no healing" and "no progression of the lesion and no healing."[10]

There was a large degree of imprecision and inconsistency in the terms used to describe clinical outcomes. Therefore, patients were considered "Functional" if they were reported as "functional," "able to perform activities of daily living," "having a full range of motion," or with "no functional restrictions." "Mild and moderate functional derangement (FD)" were considered when similar terms were found.[8] "Severe FD" was considered for terms such as "bladder and bowel dysfunction,"[19] "post-operative absent bowel and bladder function,"[20] or "patient's functional ambulation which was markedly compromised."[21]

Pain severity was categorized into "mild," "moderate," or "no pain," "Recurrence" was considered as a reappearance of the tumor in the radiographs at least 6 weeks after intervention. Terms such as "disease free," "tumor free," or "no recurrence seen" were considered as "no recurrence."

The information available on radiological and clinical outcomes was summarized according to the treatment method. Whenever information on the variables collected was not specified in the article, it was documented as not reported (NR).

The quality and study design of the articles included in this systematic review did not allow a meta-analysis. Therefore, summary measures, synthesis of results, risk of bias across studies, publication bias, and treatment effect and heterogeneity assessment could not be performed. Given the low level of evidence available, a formal quality assessment was not considered necessary.

Individual patient data were aggregated according to treatment received and descriptive statistics were applied using "StataCorp 2017."

RESULTS

A total of 143 records were identified through database searching and cross-referencing retrieved articles. After removing duplicates, 123 records were screened for eligibility [Figure 1]. After reviewing the title and abstracts, 55 records were excluded from the study. From the remaining 68 records, full-text articles were reviewed for eligibility, and 19 were omitted. Among the 19 excluded articles, 16 did not report any outcomes for specific treatment of pelvic ABC and three had a follow-up of <1 year.

The final number of studies included in the systematic review was 49, of which 29 were case reports and 20 were case series, spanning from 1979 to 2017. From the 49 studies, data were extracted from 194 patients into an excel sheet.

Treatment groups and patients included per study

Many treatment options were identified and grouped into five major treatment groups: observation, adjuvant, minor surgery, major surgery, and combined. These major treatment groups were further subdivided into 11 distinct treatment subgroups. The number of studies and patients included in each treatment group are described in [Table 1]. Some studies appeared in more than one treatment group.

Within the five major treatment groups, more than half of the patients, 113/194 (58%), were treated either with adjuvant 57/194 (29%) or minor surgery 56/194 (29%). In contrast, observation was the least used treatment accounting for only 13/194 (7%) patients.

Looking at treatment subgroups, most patients were treated with curettage 45/194 (23%) followed by SAE 38/194 (20%). Among the latter, 18 (47%) patients had only one SAE session, 10 (26%) patients had two, and further, ten patients had three sessions of SAE. Diverse biomaterials such as gel foam, latex particles, biospheres, isobutyl 2 cyanoacrylate, polyvinyl alcohol, and stainless-steel coils were used for the embolization.

Marginal excision was the third most commonly used treatment, 27/194 (14%), whereas wide excision was used only in five cases. The terms marginal and wide are derived from the "Enneking classification of surgical margins," which is a standard reported measurement of surgical margins in osseous and soft-tissue tumor surgery.[22] Various materials have been described to fill the bony voids after major or minor surgery, such as autologous bone grafts, allografts, bone cement, bioactive glass, plexur M biocomposite, arthrodesis with plating, and vascularized fibular grafts.

The combined treatment groups included any type of surgery along with either SAE, sclerotherapy, or radiotherapy. The combined group of minor surgery + adjuvant was the fourth most used treatment (26/194). Most of these patients received SAE as a neoadjuvant treatment.

Extended curettage was used in 11/194 cases and reportedly done through high-speed burr, phenol, or ethanol washes. Sclerotherapy (10/194 cases) was either performed percutaneously or through a mini-open approach under fluoroscopy and general anesthesia. The sclerosants used were ethibloc, demineralized bone matrix combined with autologous bone graft, 32 P chromic phosphate, and polidocanol. The least used methods included radiotherapy (5/194), wide excision (5/194), and SAE with sclerotherapy (4/194). Radiotherapy has been used as a neoadjuvant but also as an adjuvant and treatment for recurrence. The maximum dosage received by any patient was 6000 Gy (Gray).

Demographics, radiological, and functional outcomes were summarized according to the treatment subgroup.

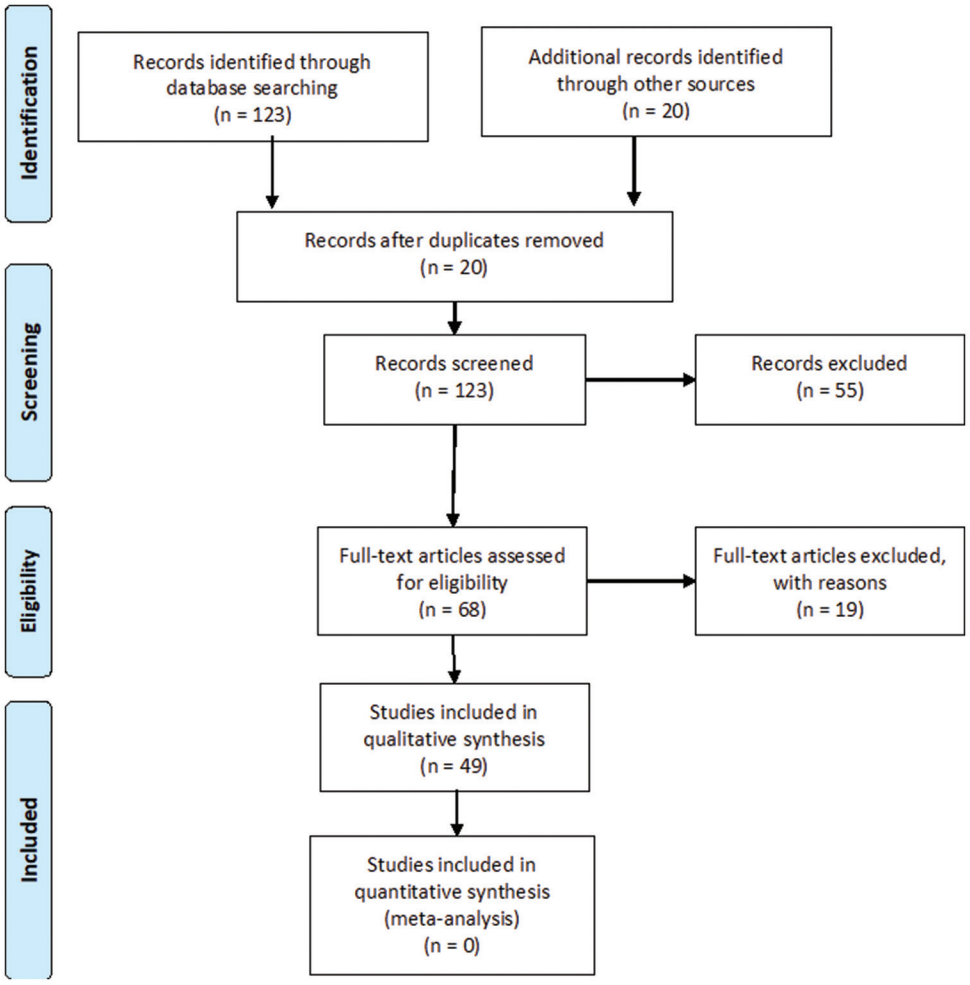

Figure 1: Flow diagram to explain the methodology of the systematic review of pelvic aneurysmal bone cyst.

Demographics

Patient demographics per treatment subgroup are described in [Table 2]. Of the 194 patients, 91 (47%) were female, 101 (52%) were male, and gender was NR for two (1%) patients. The overall median age was 15 years, ranging from 1.5 to 64 years old. The median age among the different treatment subgroups was very similar and within the second decade of life except for the wide excision group.

The mean time from the start of the symptoms to diagnosis was 10 months, ranging from 2 weeks to 10 years. The median follow-up was 3 years, with the longest follow-up of 53 years in a 10-year-old with stage 2 ABC in the sacrum treated with extended curettage and radiotherapy.[8]

Tumor characteristics

Main tumor characteristics in terms of size, anatomic location within the pelvis, and Enneking staging according to treatment group and subgroup are described in [Table 3]. Only 8 (4%) patients presented with a recurring ABC; the rest were all primary *de novo* lesions.

The information on tumor size for individual patients was available for only 50/194 patients (25%). However, for 22 (11%) patients, tumor size was described in categories of less than, up to, or >5 cm.[18] The minimum reported tumor size was 3 cm and the maximum 30 cm. The treatment groups with an average size above 15 cm were all surgical (either as a standalone treatment or combined with adjuvant therapy). In contrast, tumors in the observation group were, on average 4.5 cm and were followed up with a median of 2.8 years.

Most tumors (126/194) were located within one pelvic zone, with a majority of them in the ischiopubic area (zone 3). Only 13/194 occupied three zones: ilium, acetabulum, and ischiopubic area in all except one case (zones 1 + 2 + 3). The most frequent combination for tumors within two zones was acetabulum and ischiopubic area (zones 2 + 3), followed by ilium and acetabulum (1 + 2), and ilium and ischiopubic

Table 1: Studies and total number of patients included in each treatment group.

Treatment groups	Subgroups	No. Of studies	Studies included in the systematic review	Publication year	No. of pts[a] per study	Total no. of pts[a]
Observation	-	5	McQueen[43]	1985	2	13
			Capanna *et al.*[19]	1986	3	
			Cottalorda *et al.*[10]	2005	3	
			Louahem[44]	2012	4	
			Huschild[29]	2016	1	
Adjuvant	SAE[b]	11	Wallace *et al.*[31]	1979	1	38
			Misasi[45]	1982	2	
			Murphy[46]	1982	1	
			Keller[47]	1983	2	
			Cisneros[48]	1985	1	
			De Cristofaro *et al.*[30]	1992	5	
			Rossi *et al.*[17]	2010	22	
			Rossi[49]	2012	1	
			Doss[50]	2014	1	
			Do Brito *et al.*[51]	2015	1	
			Milan[52]	2016	1	
	Sclerotherapy	5	Flappa[33]	2002	1	10
			Dubios[34]	2003	1	
			Docquier and Delloye[24]	2005	2	
			Bush[53]	2010	1	
			Brosjo *et al.*[35]	2013	5	
	Radiotherapy	2	Capanna *et al.*[19]	1986	4	5
			Elsayad *et al.*[25]	2017	1	
	SAEb+sclerotherapy	4	Dubois *et al.*[34]	2003	1	4
			Docquier and Delloye[24]	2005	1	
			Bush[53]	2010	1	
			Simm[54]	2013	1	
Minor surgery	Curettage	9	Capanna *et al.*[19]	1986	11	45
			Abdullah[28]	1986	1	
			Papagelopoulos *et al.*[8]	2001	15	
			Cottalorda *et al.*[10]	2005	9	
			Brastianos *et al.*[18]	2009	2	
			Puri *et al.*[20]	2009	1	
			Ozdemir[55]	2011	1	
			Huschild[29]	2016	4	
			Elsayad *et al.*[25]	2017	1	
	Extended curettage	4	Khan[56]	2010	1	11
			Kim[57]	2014	1	
			Novais *et al.*[9]	2014	8	
			Hetaimish[58]	2016	1	
Major surgery	Marginal excision	6	Capanna *et al.*[19]	1986	3	27
			Malghem[59]	1989	1	
			Papagelopoulos *et al.*[8]	2001	17	
			Huang[60]	2004	1	
			Brastianos *et al.*[18]	2009	3	
			Rao[61]	2013	2	
	Wide excision	5	Papagelopoulos *et al.*[8]	2001	1	5
			Honl *et al.*[37]	2003	1	
			Yu *et al.*[36]	2007	1	
			Puri[20]	2009	1	
			Sharifah *et al.*[22]	2011	1	

(Contd...)

Table 1: *(Continued).*

Treatment groups	Subgroups	No. Of studies	Studies included in the systematic review	Publication year	No. of pts[a] per study	Total no. of pts[a]
Combined	Adjuvant+minor surgery	13	Wallace *et al.*[31]	1979	1	26
			Dick[60]	1979	1	
			Capanna *et al.*[19]	1986	2	
			Delloye *et al.*[23]	1996	1	
			Papagelopoulos *et al.*[8]	2001	2	
			Wathiong[61]	2003	1	
			Docquier and Delloye[24]	2005	2	
			Cottalorda *et al.*[10]	2005	3	
			Yildirim[64]	2007	2	
			Brastianos *et al.*[18]	2009	4	
			Novais *et al.*[9]	2014	5	
			Rossi[65]	2015	1	
			Syvanen[66]	2017	1	
	Adjuvant+major surgery	6	Cheah[67]	1999	1	10
			Papagelopoulos *et al.*[8]	2001	5	
			Pogoda[68]	2003	1	
			Brastinos[18]	2009	1	
			van de Luijtgaarden[26]	2009	1	
			Sobeai[39]	2015	1	

[a]Pts: patients, [b]SAE: Selective arterial embolization

Table 2: Patient characteristics in each treatment group.

Treatment groups	Subgroups	No. of pts[a]	Median age (min-max) yrs[b]	Gender *n* (%)			Median follow-up (min–max) yrs[b]
				Female	Male	NR[c]	
Observation	-	13	13 (10.5–46)	5 (38)	8 (62)	-	2.8 (0.4–11)
Adjuvant	SAE[d]	38	16 (3–33)	18 (47)	20 (53)	-	1.75 (0.6–8)
	Sclerotherapy	10	13.5 (5–28)	5 (50)	5 (50)	-	1.5 (0.9–4.3)
	Radiotherapy	5	15 (8–22)	1 (20)	4 (80)	-	7 (2.1–14)
	SAE[d]+Sclerotherapy	4	13.5 (8–22)	1 (25)	2 (50)	1 (25)	3.35 (1.5–5.6)
Minor surgery	Curettage	45	15 (1.5–64)	18 (40)	27 (60)	-	5 (1.2–45)
	Extended curettage	11	13.9 (5–17.5)	5 (45)	6 (55)	-	6 (0.6–19.8)
Major surgery	Marginal excision	27	14 (6–60)	13 (48)	14 (52)	-	7 (1–42)
	Wide excision	5	21 (15–51)	3 (60)	2 (40)	-	11 (3–19)
Combined	Adjuvant+minor surgery	26	13.5 (4–38)	17 (65)	8 (31)	1 (4)	4.5 (0.6–24)
	Adjuvant+major surgery	10	16.5 (10–57)	5 (50)	5 (50)	-	5.25 (1–53)

[a]pts: Patients, [b]yrs.: years, [c]NR: Not reported, [d]SAE: Selective arterial embolization. Note: Rossi *et al.*[17] grouped the age of patients as<10, 10–20, >20 yrs. Therefore, these data were excluded when calculating the median age for each treatment group

(zones 1 + 3). Tumors involving both the ilium and sacrum (zones 1 + 4) were found only in four cases.

The Enneking staging divides benign bone tumors into three categories: latent (stage 1), active (stage 2), and aggressive (stage 3).[23] Over 50% of the cases were Enneking stage 3, and 30% were stage 2. The staging was not mentioned for 30 (15%) patients.

Outcomes

Radiological and clinical outcomes per treatment group are detailed in [Table 4]. Given that the radiological and functional outcomes reported in the retrieved studies were remarkably inconsistent, heterogenous, and, on occasions, extremely vague, we tried to categorize the outcomes following our best interpretation and understanding of what was reported by the authors. Only one study[9] described the outcomes using the musculoskeletal tumor rating scale, Toronto extremity salvage score, and the 36-item short-form health survey.

Our primary outcome focused on ossification. In total, 150 patients (77%) had complete ossification, 19 patients (10%) had partial, and five patients (2%) had no evidence of ossification. The ossification status was unavailable in

Table 3: Tumor characteristics in each treatment group.

Treatment groups	Subgroups	No. of pts[a]	*Mean size cm[b]	Pelvic zone location *n* (%)										Enneking staging *n* (%)			
				Single zone				Two zones				Three zones					
				1[c]	2[d]	3[e]	4[f]	1+2	1+3	1+4	2+3	1+2 + 3	1+2 + 4	1	2	3	NR[g]
Observation	-	13	4.5	1 (8)	1 (8)	5 (38)	-	-	1 (8)	-	1 (8)	4 (30)	-	1 (8)	4 (31)	7 (53)	1 (8)
Adjuvant	SAE[h]	38	7.3	12 (32)	2 (5)	15 (40)	7 (18)	-	-	2 (5)	-	-	-	-	16 (42)	12 (32)	10 (26)
	Sclerotherapy	10	7.68	1 (10)	1 (10)	3 (30)	2 (20)	-	1 (10)	-	2 (20)	-	-	-	1 (10)	9 (90)	-
	Radiotherapy	5	NR	-	-	-	-	-	3 (60)	1 (20)	-	1 (20)	-	-	-	-	5 (100)
	SAE[h]+ Sclerotherapy	4	7.83	-	-	-	2 (50)	-	1 (25)	-	-	1 (25)	-	-	-	1 (25)	3 (75)
Minor surgery	Curettage	45	22.5	8 (17)	2 (4)	8 (17)	11 (24)	2 (4)	4 (8)	1 (2)	4 (8)	5 (11)	-	1 (25)	15 (33)	23 (51)	6 (13)
	Extended curettage	11	11	1 (9)	-	7 (63)		3 (27)	-	-	-	-	-	-	8 (72)	3 (27)	-
Major surgery	Marginal excision	27	10.8	2 (7)	-	4 (15)	8 (30)	2 (7)	1 (10)	-	10 (37)	-	-	-	7 (26)	19 (70)	1 (4)
	Wide excision	5	18.5	-	1 (20)	-	1 (20)		1 (20)	-	1 (20)	-	1 (20)	-	-	4 (75)	1 (25)
Combined	Adjuvant+ minor surgery	26	7.77	2 (8)	2 (8)	5 (19)	6 (23)	5 (19)	2 (8)	-	3 (12)	1 (3)	-	-	6 (23)	17 (65)	3 (12)
	Adjuvant+ major surgery	10	21.5	1 (10)	-	1 (10)	4 (40)	3 (30)	-	-	1 (10)	-	-	-	1 (10)	9 (90)	-

[a]pts: Patients, [b]Size: mean size of the tumor in centimeters using the maximum dimension reported, [c]1=ilium, [d]2=acetabulum, [e]3=ischiopubic, [f]4=sacrum, [g]NR: Not reported, [h]SAE: selective arterial embolization. *Data for tumor size were available for only 50/194 patients

Table 4: Radiological and clinical outcomes by treatment group.

Treatment	Subgroups	No. of pts[a]	Ossificationn (%)				Functional outcomen (%)					Painn (%)				Recurrencen (%)		
			Yes	Partial	No	NR[b]	Functional[c]	Mild FD[d]	Moderate FD[e]	Severe FD[f]	NR[b]	No pain	Mild	Moderate	NR[b]	Yes	No	NR[b]
Obs[g]	-	13	9 (69)	3 (23)	-	1 (8)	12 (92)	-	-	-	1 (8)	12 (92)	-	1 (8)	1 (8)	-	12 (92)	1 (8)
Adjuvant	SAE[h]	38	28 (74)	7 (18)	3 (8)	-	14 (37)	-	-	-	24 (63)	14 (37)	-	-	24 (63)	1 (3)	37 (97)	-
	Sclerotherapy	10	10 (100)	-	-	-	8 (80)	-	-	-	2 (20)	8 (80)	-	-	2 (20)	-	9 (90)	1 (10)
	Radiotherapy	5	4 (80)	1 (20)	-	-	1 (20)	-	2 (40)	1 (20)	1 (20)	2 (40)	2 (40)	1 (20)	-	1 (20)	4 (80)	-
	SAE[h]+ Sclerotherapy	4	4 (100)	-	-	-	1 (25)	-	-	-	3 (75)	1 (25)	-	-	3 (75)	-	4 (100)	-
Minor surgery	Curettage	45	34 (75)	5 (11)	1 (2)	5 (11)	40 (88)	2 (4)	-	2 (4)	1 (2)	34 (75)	6 (13)	1 (2)	4 (8)	9 (20)	30 (66)	6 (13)
	Extended curettage	11	11 (100)	-	-	-	8 (72)	1 (9)	1 (9)	-	1 (9)	3 (27)	-	-	8 (72)	1 (9)	9 (81)	1 (9)
Major surgery	Marginal excision	27	23 (85)	-	-	4 (14)	22 (81)	3 (11)		1 (3)	1 (3)	19 (70)	4 (14)	-	4 (14)	2 (7)	24 (88)	1 (3)
	Wide excision	5*	3 (60)	-	-	1 (20)	2 (40)	1 (20)	-	-	1 (20)	1 (20)	1 (20)	1 (20)	1 (20)	1 (20)	2 (40)	1 (20)
Combined	Adjuvant+ minor surgery	26	16 (61)	3 (11)	-	7 (26)	15 (57)	3 (11)	2 (7)	-	6 (23)	12 (46)	1 (3)	1 (3)	12 (46)	3 (11)	19 (73)	4 (15)
	Adjuvant+ major surgery	10*	8 (80)	-	1 (20)	-	7 (70)	1 (10)	1 (10)	-	-	6 (60)	2 (20)	-	1 (10)	3 (30)	6 (60)	-

[a]pts: patients, [b]NR: Not reported, [c]Functional: Having full range of motion/activities of daily living/no functional restriction, [d]Mild FD: Mild functional derangement, [e]Moderate FD: Moderate functional derangement, [f]Severe FD: Severe functional derangement, [g]obs: Observation, [h]SAE: Selective arterial embolization. *One patient died. Therefore, the sum of the outcomes is less than the total number of patients in these categories

20/194 cases: in 18 patients (9%), it was NR, and two patients (1%) died during or shortly after surgery.

Clinical outcomes related to function, pain, and recurrence. Patients were reported as functional in 140 (72%) cases. However, mild FD was seen in 11 patients (6%), moderate in 6 (3%), severe in 4 (2%), and in 41 (21%), the functional outcome was NR. At the end of the follow-up, 112 patients (58%) reportedly had no pain, whereas mild pain was mentioned in 16 (8%) patients, moderate pain in 5 (2%) patients, and 50 patients (30%), the pain was NR.

Overall, the incidence of recurrence was low. Recurrence was reported in 21 (10.8%) patients and NR in 15 patients (8%), whereas the remaining patients had no recurrence at the time of the last follow-up. The longest time to recurrence was 6 years in a 13-year-old male patient with an ABC in the pubis extending to the acetabulum treated with an extended curettage.[8] Recurrence was treated mostly by curettage or excision and autologous bone grafting. Few recurrent cases were managed with scleroembolic treatments instead of surgery[24] and three cases were treated with radiotherapy.[22,25] Curettage had a relatively high number of recurrences compared to the other treatments. However, the unbalanced group size and a low number of patients in some subgroups preclude a proper statistical analysis.

The number of reported complications is low. Hemorrhage was reported in six (3%) patients who were treated with surgery (five patients) and sclerotherapy (one patient). Of these, one patient died secondary to massive bleeding. Neurological complications were described in 13 (7%) patients. These include hypoesthesia, bowel and bladder dysfunction or residual loss of bowel, bladder control, neuroma formations, and involvement of sacral roots, sciatic nerve, obturator nerve, and caudal sac. Eleven (6%) patients with neurological complications had ABC located in zone 4 (sacral), while the remaining two were in zones 2 and 3. One patient with a tumor in zones 2 and 3 had partial encasement of the bladder with the ABC mass resulting in subtotal cystectomy. Limb length discrepancy (0.6 – 3cm) was reported in four patients (2%) after marginal resection of ABC. Curettage in the sacrum (pelvic zone 4) was performed in 12 (26%) cases and had frequent reports of hemorrhage, bowel injury, dural tear, and loss of bowel bladder control. Nine out of 21 (42%) patients had complications after treatment for recurrence. Capanna *et al.* reported three cases, in which the primary treatment was radiotherapy or a combination of radiotherapy with surgery. Recurrence was observed and treated with curettage or radiotherapy. These patients were left with residual complications of crural nerve palsy, quadriceps, psoas deficit, and femoral head necrosis.[20]

Two patients died of treatment-related complications in this series of 194 cases reported in the literature. A 51-year-old female with a 25 × 25 × 20 cm ABC in the right ilium extending into the ischium. The patient was treated surgically and died of profuse perioperative bleeding.[26] The second case was a 48-year-old female with a 27.6 × 22.4 × 15.9 cm lesion in the left ilium with a history of embolization, sclerotherapy, and repeated surgery. The authors performed pre-operative embolization and percutaneous intralesional alcohol injection. This was followed by staged near-total resection combined with cryosurgery and off-label systemic treatment with bevacizumab, a monoclonal anti-vascular endothelial growth factor antibody, to reduce bleeding. The patient developed clinical heart failure, prompting the discontinuation of bevacizumab, followed by a rapid regrowth of the pelvic lesion. After seven surgical sessions, the patient's condition no longer allowed further interventions and she was treated with radiotherapy. The patient died 2 years after ABC was first diagnosed.[27]

ABC in the pelvis has also been described in two pregnant women. A pelvic bone lesion in pregnancy can be difficult for the obstetrician to determine the effect of parturition on the pelvic bone.[28] The first reported case was in 1986 of a right iliac ABC measuring 15 cm in a 19-year-old primigravida. At the time of the elective caesarian section, the mass was excised through a separate lumbar approach. No recurrence or complication was reported.[29] The second reported case was a 6.1 × 2.9 × 6 cm lesion in the left ilium of a 26-year-old primigravida who had a successful elective cesarian section, and the ABC was observed until the last follow-up at 3 months.[28]

We, further, analyzed tumor characteristics and clinical and radiological outcomes according to their pelvic zone location and the results are summarized in [Table 5]. While zone 3 was overall the most common tumor location 48/194 (25%), when taking into consideration tumors with overlapping zones as well, most patients 74/194 (38%) had an affection of zone 1 – ilium; of these 14 had partial ossification and the functional outcome was NR in half of them. Tumors in zone 4 sacrum had more neurological complications reported, such as bladder and bowel dysfunction.[19] Moderate residual pain was mentioned in 4/194 patients located in zones 4, 2 + 3, and 1 + 2 + 3. Moreover, severe FD reported in 4/194 were located in zone 4 (3 cases) and zone 1 + 3 (1 case).

DISCUSSION

A pelvic ABC is a treatment challenge. Most cases have been managed with individualized treatment plans tailored to their size, stage, location, behavior, available clinical resources, and surgical expertise. In this systematic literature review, we were able to successfully categorize these varied treatments for 194 patients of pelvic ABC in five groups and 11 sub-groups. This is not only a useful overview of existing therapeutic modalities but also suggests the variability that exists in choosing the best treatment modality.

Table 5: Tumor characteristics and outcomes by pelvic zones.

Pelvic Zones	No. of pts[a]	#Mean size (cm)[b]	Enneking stage *n* (%)				Ossification *n* (%)				Functional outcome *n* (%)					Pain *n* (%)				Recurrence *n* (%)		
			1	2	3	NR[c]	Yes	Partial	No	NR[c]	Functional[d]	Mild FD[e]	Moderate FD[f]	Severe FD[g]	NR[c]	No pain	Mild	Moderate	NR[c]	Yes	No	NR[c]
1*	28	10	1 (4)	11 (39)	10 (36)	6 (21)	15 (53)	10 (35)	2 (7)	-	14 (50)	-	-	-	13 (46)	12 (42)	2 (7)	-	13 (46)	6 (21)	20 (71)	1 (3)
2	9	9.06	-	2 (22)	6 (67)	1 (11)	7 (77)	1 (11)	-	1 (11)	8 (88)	-	-	-	1 (11)	7 (77)	-	-	2 (22)	-	9 (100)	-
3	48	8.2	1 (2)	20 (41)	19 (40)	8 (17)	41 (85)	4 (8)	2 (4)	1 (2)	36 (75)	1 (2)	1 (2)	-	10 (20)	31 (64)	-	-	17 (35)	3 (6)	40 (83)	5 (10)
4	41	11.6	-	9 (22)	26 (63)	6 (15)	28 (68)	-	-	13 (31)	23 (56)	6 (15)	-	3 (7)	9 (21)	18 (37)	5 (12)	2 (4)	16 (39)	6 (15)	35 (85)	-
1+2	15	7.5	-	5 (33)	10 (67)	-	13 (86)	-	-	1 (6)	9 (60)	4 (26)	1 (6)	-	1 (6)	7 (46)	2 (13)	-	6 (40)	2 (13)	13 (87)	-
1+3*	14	15.5	-	4 (29)	10 (71)	-	11 (78)	1 (7)	1 (7)	-	8 (57)	-	1 (7)	1 (7)	3 (21)	8 (57)	2 (14)	-	3 (21)	3 (21)	9 (64)	1 (7)
1+4	4	NR[c]	-	1 (25)	2 (50)	1 (25)	3 (75)	1 (25)	-	-	3 (75)	-	-	-	1 (25)	3 (75)	1 (25)	-	-	-	4 (100)	-
2+3	22	7.75	-	5 (23)	17 (77)	-	21 (95)	1 (5)	-	-	21 (95)	-	1 (5)	-	-	18 (81)	3 (13)	1 (5)	-	1 (5)	20 (90)	1 (5)
1+2 + 3	12	11	-	2 (17)	10 (83)	-	8 (66)	1 (8)	-	3 (25)	8 (66)	-	1 (8)	-	3 (25)	7 (58)	1 (8)	1 (8)	3 (25)	1 (8)	7 (58)	4 (33)
1+2 + 4	1	NR[c]	-	-	1 (100)	-	1 (100)	-	-	-	-	-	-	-	1 (100)	1 (100)	-	-	-	-	-	1 (100)

[a]pts: Patients, [b]Mean size of the tumor in centimeters using the maximum dimension reported, [c]NR: No reported, [d]Functional: Having full range of motion/activities of daily living/no functional restriction, [e]Mild FD: Mild functional derangement, [f]Moderate FD: Moderate functional derangement, [g]Severe FD: severe functional derangement. *One patient died. Therefore, the sum of the outcomes is less than the total number of patients in these categories, [#]Data for tumor size were available for only 50/1

We could identify only 194 patients of pelvic ABC, fulfilling our inclusion criteria since 1979, which emphasizes the rarity of the problem. In our study, the gender distribution of pelvic ABC had a slight male predominance in 52% of the cases. The median age was 15 years, although the range was quite wide, from 1.5 years to 65 years. Most lesions grow significantly, become stage 3, and remain asymptomatic before being diagnosed. Among the 194 reported treated cases in the literature, 60% showed complete radiological ossification, 72% were reported functional, and 10.8% had a recurrence. These results indicate that most pelvic ABCs have a good prognosis in general.

Curettage with bone grafting is the most preferred treatment method, followed by SAE either alone or in combination with surgery. In the curettage treatment group, 75% had radiological ossification with no pain and 88% were "functional" after treatment.[8,9,19,20,30] It was used in all pelvic zones and for any tumor stage. The maximum reported size for a lesion treated with curettage alone was 30 × 30 × 20 cm in zone 4 in a 14-years-old.[25]

SAE is described in the literature as a simpler, cheap, easily repeatable, less invasive, or less risky procedure. It has been used in patients at extreme ages, the youngest being 1.5 and the oldest 64 years. Surgery and radiotherapy were the main treatment modalities for ABC until the introduction of SAE.[31] The earliest records of SAE for pelvic ABC come from Wallace *et al.* in two patients. They reported a 15% reduction in size and pain-free status after 1 year in one 27-year-old female patient with an ABC in the ilium. The second patient, a 38-year-old male, was reported to have surgery and cordotomy followed by SAE. The patient remained pain-free at 5 years follow-up but had a recurrence later.[32]

Varshney *et al.* did a comparative study to determine the healing rate and functional score between two groups treated with sclerotherapy (polidocanol) or intralesional curettage. They concluded that "although the healing rates were similar, we found higher rates of clinically important complications, worse functional outcomes, and higher hospital burden associated with intralesional excision." They called it a preliminary study and recommended larger studies to confirm their findings.[33] In this review, a comparison of the two groups was not possible due to a large discrepancy in patient number 45 in curettage versus 10 in the sclerotherapy group. Moreover, due to the poor quality of reporting outcomes, the reliability of the results is questionable.

In most recent studies reviewed, scleroembolic procedures are used as adjuncts or combined with major surgical procedures in large pelvic tumors.[33-36] Papagelopoulos *et al.* set five cm as a cut-off for performing a curettage or excisional surgery.[8] We observed in the systematic review that tumors >15 cm were all treated surgically either alone or combined with adjuvants. However, we cannot say if this is the best treatment in such cases. Following an excisional surgery, some major mechanical complications are reported, such as hip dislocation/subluxation, protrusio acetabuli, and limb length discrepancy.[8] However, according to the reports, the key to overcoming these issues is to get a near anatomic reconstruction of the pelvic defects. At present, tricortical iliac crest grafts, vascularized or non-vascularized fibular grafts, allografts, custom-made mega prosthesis, extracorporeal irradiated pelvic bone, and re-implantation have all been described.[19,21,37-39] Zidzislaw *et al*. predicted that the future for pelvic defects is a statistical shape model reconstruction method using a mirroring approach.

Since the anatomic location of the tumor influences the treatment strategy and prognosis, we analyzed the results by pelvic zone. Patients with tumors in zone 4 accounted for most of the neurological complications reported, whereas half of the cases with hemorrhage, including a deadly case, were located in zone 1. In their series of 13 pelvic ABCs in children, Novais *et al*. observed higher recurrence rates in open physis and suggested that destructive lesions in triradiate cartilage can lead to long-term complications.[9] In our results, we observed that 3/4 of patients with limb length discrepancy had their ABCs near the triradiate cartilage, that is, zones 2 and 3. In summary, the particular neighboring structures and their involvement require modified treatment methods for better outcomes, as Papagelopolus concluded.[8]

The minimum average tumor size per treatment group was 4.5 cm (observation), whereas the maximum average size was 22.5 cm (curettage). However, when the results are grouped by pelvic zones, the average diameter ranges only from 7.5 to 15.5 cm. This might suggest that in addition to the anatomic location, the tumor size also has some influence on the treatment method since the average size by pelvic zone is similar. However, since tumor size was only available for 25% of patients, these results should be interpreted with great caution.

Denosumab is a human monoclonal antibody that prevents the cytokine receptor activator of nuclear factor-kappa B ligand from activating the RANK receptor of osteoclasts, inhibiting osteoclast function. Denosumab is highly effective in giant cell tumors of bone (GCT), and therefore, similar effects in principle could be hoped for in ABC, which has distinct similarities to GCT. Up to now, no protocol or treatment recommendation for the use of denosumab in ABC exists. Dürr *et al*., while concluding off-label use of Denosumab in six patients of ABC (out of which two were sacral, one pelvic in location), suggest it as an adjuvant option in anatomically challenging locations and local recurrence. However, they reported severe rebound hypercalcemia in young patients.[40] Since this systematic review was compiled in September 2017; these reports are not part of the results.

The limitations of our systematic literature review are mainly due to the low evidence found, which was mostly restricted to retrospective case reports and case series. This was, further,

hampered by the heterogeneous reporting and inconsistent outcome measurements, which precluded a meta-analysis. The systematic review was performed in September 2017, so reports published after that are not included in this study.

CONCLUSION

Pelvic ABC has been documented in medical literature as case reports or case series and with heterogeneous reporting. In this systematic literature review, individual patient data were extracted to give an overview regarding functional and radiological outcomes by treatment modality.

Although mostly benign, pelvic ABC has unpredictable behavior. A clear trend is being adopted toward using neoadjuvant scleroembolic treatments alone or combined with surgery for better radiological and functional outcomes.

RECOMMENDATIONS

We encourage reporting of surgical case reports following a standard reporting guideline like the SCARE statement (http://www.scareguideline.com/) to strengthen the quality of reporting.[41,42]

ACKNOWLEDGMENT

The present research work was supported by AOTrauma in the form of a clinical research fellowship at AOCID granted to the first author. We are grateful to the AOCID staff, and in particular to Vasiliki Kalampoki, for her help in data analysis. We also acknowledge the contribution of Prof Syed Muhammad Awais, Dean Faculty of Health Sciences, University of Punjab, Lahore, Pakistan, for his generous sharing of knowledge, constant support, and for being a source of inspiration.

ETHICAL APPROVAL

An ethics statement is not applicable, because this study is based exclusively on published literature.

AUTHORS' CONTRIBUTIONS

The authors confirm contribution to the paper as follows: study conception and design, draft manuscript preparation: AS, ER and AHC. AHC and AS mainly performed the analysis and interpretation of results. Data collection was done by AS. All authors have critically reviewed and approved the final draft and are responsible for the manuscript's content and similarity index.

DECLARATION OF PATIENT CONSENT

Patients consent is not applicable, because this study is based exclusively on published literature.

FINANCIAL SUPPORT AND SPONSORSHIP

This study did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

CONFLICTS OF INTEREST

There are no conflicting relationships or activities.

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;23:6756-62.
2. Jaffe HL, Lichtenstein L. Solitary unicameral bone cyst: With emphasis on the roentgen picture, the pathologic appearance and the pathogenesis. Arch Surg 1942;4:1004-25.
3. Jaffe HL. Aneurysmal bone cyst. Bull Hosp Joint Dis 1950;11:3-13.
4. De Dios AM, Bond JR, Shives TC, McLeod RA, Unni KK. Aneurysmal bone cyst. A clinicopathologic study of 238 cases. Cancer 1992;69:2921-31.
5. Szendroi M, Cser I, Konya A, Renyi-Vamos A. Aneurysmal bone cyst. A review of 52 primary and 16 secondary cases. Arch Orthop Trauma Surg 1992;111:318-22.
6. Leithner A, Windhager R, Lang S, Haas OA, Kainberger F, Kotz R. Aneurysmal bone cyst. A population based epidemiologic study and literature review. Clin Orthop Relat Res 1999;363:176-9.
7. Zehetgruber H, Bittner B, Gruber D, Krepler P, Trieb K, Kotz R, *et al*. Prevalence of aneurysmal and solitary bone cysts in young patients. Clin Orthop Relat Res 2005;439:136-43.
8. Schreuder HW, Veth RP, Pruszczynski M, Lemmens JA, Koops HS, Molenaar WM. Aneurysmal bone cysts treated by curettage, cryotherapy and bone grafting. J Bone Joint Surg Br 1997;79:20-5.
9. Kransdorf MJ, Sweet DE. Aneurysmal bone cyst: Concept, controversy, clinical presentation, and imaging. AJR Am J Roentgenol 1995;164:573-80.
10. Papagelopoulos PJ, Choudhury SN, Frassica FJ, Bond JR, Unni KK, Sim FH. Treatment of aneurysmal bone cysts of the pelvis and sacrum. J Bone Joint Surg Am 2001;83:1674-81.
11. Novais EN, Zimmerman AK, Lewallen LW, Rose PS, Sim FH, McIntosh AL. Functional outcomes and quality of life following surgical treatment of aneurysmal bone cysts of the pelvis in children. J Child Orthop 2014;8:281-8.
12. Cottalorda J, Chotel F, Kohler R, de Gauzy JS, Louahem D, Lefort G, *et al*. Aneurysmal bone cysts of the pelvis in children: A multicenter study and literature review. J Pediatr Orthop 2005;25:471-5.
13. Saeed A, Chong AH, Rometsch E, Awais SM. A rare case of huge aneurysmal bone cyst of the pelvis. J Musculoskelet Surg Res 2021;5:201-5.
14. Liberati A, Altman DG, Tetzlaff J, Mulrow C, Gotzsche PC, Ioannidis JP, *et al*. The PRISMA statement for reporting systematic reviews and meta-analyses of studies that evaluate healthcare interventions: Explanation and elaboration. BMJ 2009;339:b2700.
15. Moher D, Liberati A, Tetzlaff J, Altman DG, PRISMA Group.

Preferred reporting items for systematic reviews and meta-analyses: The PRISMA statement. PLoS Med 2009;6:e1000097.
16. Enneking WF, Dunham WK. Resection and reconstruction for primary neoplasms involving the innominate bone. J Bone Joint Surg Am 1978;60:731-46.
17. Enneking WF. A system of staging musculoskeletal neoplasms. Clin Orthop Relat Res 1986;204:9-24.
18. Rossi G, Rimondi E, Bartalena T, Gerardi A, Alberghini M, Staals EL, *et al*. Selective arterial embolization of 36 aneurysmal bone cysts of the skeleton with N-2-butyl cyanoacrylate. Skeletal Radiol 2010;39:161-7.
19. Brastianos P, Gokaslan Z, McCarthy EF. Aneurysmal bone cysts of the sacrum: A report of ten cases and review of the literature. Iowa Orthop J 2009;29:74-8.
20. Capanna R, Bertoni F, Bettelli G, Present D, Biagini R, Ruggieri P, *et al*. Aneurysmal bone cysts of the pelvis. Arch Orthop Trauma Surg(1978) 1986;105:279-84.
21. Puri A, Agarwal MG, Shah M, Srinivas CH, Shukla PJ, Shrikhande SV, *et al*. Decision making in primary sacral tumors. Spine J 2009;9:396-403.
22. Enneking WF, Spanier SS, Goodman MA. A system for the surgical staging of musculoskeletal sarcoma. Clin Orthop Relat Res 1980;153:106-20.
23. Delloye C, De Nayer P, Malghem J, Noel H. Induced healing of aneurysmal bone cysts by demineralized bone particles. A report of two cases. Arch Orthop Trauma Surg 1996;115:141-5.
24. Docquier PL, Delloye C. Treatment of aneurysmal bone cysts by introduction of demineralized bone and autogenous bone marrow. J Bone Joint Surg Am 2005;87:2253-8.
25. Elsayad K, Kriz J, Seegenschmiedt H, Imhoff D, Heyd R, Eich HT, *et al*. Radiotherapy for aneurysmal bone cysts: A rare indication. Strahlenther Onkol 2017;193:332-40.
26. Sharifah M, Nurhazla H, Suraya A, Tan S. Pelvic aneurysmal bone cyst. Biomed Imaging Interv J 2011;7:e24.
27. Van de Luijtgaarden AC, Veth RP, Slootweg PJ, Wijers-Koster PM, Kool LJ, Bovee JV, *et al*. Metastatic potential of an aneurysmal bone cyst. Virchows Arch 2009;455:455-9.
28. Elkattah R, Foulk B. A suspected pelvic aneurysmal bone cyst in pregnancy. Case Rep Obstet Gynecol 2013;2013:676087.
29. Issa AA, Amr SS, Swaiss AM. Aneurysmal bone cyst of the ilium associated with pregnancy. Eur J Obstet Gynecol Reprod Biol 1986;23:243-8.
30. Hauschild O, Ludemann M, Engelhardt M, Baumhoer D, Baumann T, Elger T, *et al*. Aneurysmal bone cyst (ABC): Treatment options and proposal of a follow-up regime. Acta Orthop Belg 2016;82:474-83.
31. De Cristofaro R, Biagini R, Boriani S, Ricci S, Ruggieri P, Rossi G, *et al*. Selective arterial embolization in the treatment of aneurysmal bone cyst and angioma of bone. Skeletal Radiol 1992;21:523-7.
32. Wallace S, Granmayeh M, deSantos LA, Murray JA, Romsdahl MM, Bracken RB, *et al*. Arterial occlusion of pelvic bone tumors. Cancer 1979;43:322-8.
33. Varshney MK, Rastogi S, Khan SA, Trikha V. Is sclerotherapy better than intralesional excision for treating aneurysmal bone cysts? Clin Orthop Relat Res 2010;468:1649-59.
34. Falappa P, Fassari FM, Fanelli A, Genovese E, Ascani E, Crostelli M, *et al*. Aneurysmal bone cysts: Treatment with direct percutaneous ethibloc injection: Long-term results. Cardiovasc Intervent Radiol 2002;25:282-90.
35. Dubois J, Chigot V, Grimard G, Isler M, Garel L. Sclerotherapy in aneurysmal bone cysts in children: A review of 17 cases. Pediatr Radiol 2003;33:365-72.
36. Brosjo O, Pechon P, Hesla A, Tsagozis P, Bauer H. Sclerotherapy with polidocanol for treatment of aneurysmal bone cysts. Acta Orthop 2013;84:502-5.
37. Yu G, Zhang F, Zhou J, Chang S, Cheng L, Jia Y, *et al*. Microsurgical fibular flap for pelvic ring reconstruction after periacetabular tumor resection. J Reconstr Microsurg 2007;23:137-42.
38. Honl M, Westphal F, Carrero V, Morlock M, Schwieger K, Hille E, *et al*. Pelvic girdle reconstruction based on spinal fusion and ischial screw fixation in a case of aneurysmal bone cyst. Sarcoma 2003;7:177-82.
39. Al Sobeai M, Al Juhani W. Pelvic aneurysmal bone cyst: Case report and literature review. Int J Surg Case Rep 2015;1:20-5.
40. Dürr HR, Grahneis F, Baur-Melnyk A, Knösel T, Birkenmaier C, Jansson V, *et al*. Aneurysmal bone cyst: Results of an off label treatment with denosumab. BMC Musculoskelet Disord 2019;20:456.
41. Agha RA, Fowler AJ, Saeta A, Barai I, Rajmohan S, Orgill DP, *et al*. The SCARE statement: Consensus-based surgical case report guidelines. Int J Surg 2016;34:180-6.
42. Jackson D, Daly J, Saltman DC. Aggregating case reports: A way for the future of evidence-based health care? Clin Case Rep 2014;2:23-4.
43. McQueen MM, Chalmers J, Smith GD. Spontaneous healing of aneurysmal bone cysts. A report of two cases. J Bone Joint Surg Br 1985;67:310-2.
44. Louahem D, Kouyoumdjian P, Ghanem I, Mazeau P, Perrochia H, L'kaissi M, *et al*. Active aneurysmal bone cysts in children: possible evolution after biopsy. J Child Orthop 2012;6:333-8.
45. Misasi N, Cigala F, Iaccarino V, Cozzolino F, Sadile F, Marasco E. Selective arterial embolisation in aneurysmal bone cysts. Int Orthop 1982;6:123-8.
46. Murphy WA, Strecker EB, Schoenecker PL. Transcatheter embolisation therapy of an ischial aneurysmal bone cyst. J Bone Joint Surg Br 1982;64:166-8.
47. Keller FS, Rosch J, Bird CB. Percutaneous embolization of bony pelvic neoplasms with tissue adhesive. Radiology 1983;147:21-7.
48. Cisneros AJ, Gomez CH, Benedicto FC. Aneurysmal bone cyst of the ischium. Int Orthop 1985;9:49-54.
49. Rossi G, Mavrogenis AF, Papagelopoulos PJ, Rimondi E, Ruggieri P. Successful treatment of aggressive aneurysmal bone cyst of the pelvis with serial embolization. Orthopedics 2012;35:e963-8.
50. Doss VT, Weaver J, Didier S, Arthur AS. Serial endovascular embolization as stand-alone treatment of a sacral aneurysmal bone cyst. J Neurosurg Spine 2014;20:234-8.
51. do Brito JS, Portela J. Selective arterial embolization for a large pelvic aneurysmal bone cyst treatment. Acta Med Port 2015;28:780-3.
52. Milan NS, Almero LP, Minguez Rey MF. Background acetabular aneurysmal bone cyst in a 7 year-old: Presentation of a case. Rev Esp Cir Ortop Traumatol 2016;60:256-9.
53. Bush CH, Adler Z, Drane WE, Tamurian R, Scarborough MT, Gibbs CP. Percutaneous radionuclide ablation of axial aneurysmal

bone cysts. AJR Am J Roentgenol 2010;194:W84-90.
54. Simm PJ, O'Sullivan M, Zacharin MR. Successful treatment of a sacral aneurysmal bone cyst with zoledronic acid. J Pediatr Orthop 2013;33:e61-4.
55. Ozdemir HM, Yurtcu M, Ogun TC. A giant recurrent aneurysmal bone cyst of the sacrum: Discussion of total resectability in a pediatric case. Turkiye Klinikleri Tip Bilimleri Dergisi 2011;31:1288-92.
56. Khan AQ, Siddiqui YS, Parameshwar A, Anwar-Sherwani MK. Aneurysmal bone cyst of pubis. A rare presentation. Saudi Med J 2010;31:317-20.
57. Kim CG, Kweon SH. Primary aneurysmal bone cyst in the iliac bone: A case report. Hip Pelvis 2014;26:202-5.
58. Hetaimish BM, Alshaya OS. Pediatric aneurysmal bone cyst in the ischial region. Saudi Med J 2016;37:799-803.
59. Malghem J, Maldague B, Esselinckx W, Noel H, De Nayer P, Vincent A. Spontaneous healing of aneurysmal bone cysts. A report of three cases. J Bone Joint Surg Br 1989;71: 645-50.
60. Huang TL, Chen WM, Chen WY, Chen TH. Huge aneurysmal bone cyst of iliac bone in a mid-aged female. J Chin Med Assoc 2004;67:99-103.
61. Rao A, Samant PD, Varshneya A. Aneurysmal bone cyst of pubic ramus: A rare entity. J Orthop Case Rep 2013;3:42-5.
62. 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;139:133-41.
63. Wathiong J, Brys P, Samson I, Maleux G. Selective arterial embolization in the treatment of an aneurysmal bone cyst of the pelvis. JBR-BTR 2003;86:325-8.
64. Yildirim E, Ersözlü S, Kirbaş I, Ozgür 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. Diagn Interv Radiol 2007;13:49-52.
65. Rossi B, Zoccali G, Marolda G, Erba F, Zoccali C. Engineering of human bone with Plexur M in acetabular roof reconstruction after curettage of a giant aggressive aneurismal bone cyst of the left emypelvis. Minerva Pediatr 2015;67:106-9.
66. Syvanen J, Nietosvaara Y, Kohonen I, Koskimies E, Haara M, Korhonen J, *et al*. Treatment of aneurysmal bone cysts with bioactive glass in children. Scand J Surg 2018;107:76-81.
67. Cheah HK, Griffin AM, White LM. Musculoskeletal images. Aneurysmal bone cyst of pelvis. Can J Surg 1999;42:411-2.
68. Pogoda P, Linhart W, Priemel M, Rueger JM, Amling M. Aneurysmal bone cysts of the sacrum. Clinical report and review of the literature. Arch Orthop Trauma Surg 2003;123:247-51.