Acta Orthop. Belg., 2016, **82**, 474-483 JSCHILD *ORIGINAL STUDY*

Aneurysmal bone cyst (ABC) : treatment options and proposal of a follow-up regime

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The aim of this study was to describe treatment options and develop a follow-up regime for the aneurysmal bone cyst, a neoplastic bone lesion with a noticeable recurrence rate. Reports of 28 patients and a mean follow-up of 42.2 months treated multidisciplinary were analysed. Data were complemented by a literature review including 790 patients. Patient age was from seven to 57 years, in line with the literature (1-69 years). Lesions most frequently affect long bones, spine and pelvis ; pain is the most common symptom. Treatment modalities vary, recurrences occurred in 26.1% in our series, rates ranged from 0-60% in the literature, with the vast majority within 2 years. With regard to the findings we propose, irrespective of treatment, a follow-up regime including clinical survey and imaging, best with MRI, at 3 months, 6 months and at half-yearly intervals within the first two and yearly within the third to fifth year.

Keywords : aneurysmal bone cyst ; symptom ; treatment ; recurrence ; follow-up.

INTRODUCTION

The aneurysmal bone cyst (ABC) is an osteolytic, hyperaemic-hemorrhagic lesion of bone which may arise de novo (primary ABC), or secondarily complicating numerous tumors and tumor-like lesions of bone (secondary ABC), such as giant cell tumor, chondroblastoma, fibrous dysplasia, solitary bone cyst or osteoblastoma (*8,13,18,23*).

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No benefits or funds were received in support of this study.
The authors report no conflict of interests.

Initially regarded as a hyperplastic lesion reactive to an unknown local process of uncertain aetiology (*20*), the primary ABC is now widely accepted as a neoplasm owing to recent findings of recurrent chromosomal rearrangements involving the USP6 gene (*20*). Malignant transformation usually to an osteosarcoma is an unlikely event, but a few cases have been reported (*14,22*).

The lesion affects all age groups, but is most common during the first two decades of life (*8,13, 18,23*) and the estimated annual incidence is 0.14 per 10^5 individuals (*16*). It comprises approximately 1 % of all bone tumors who underwent biopsy (*8, 16*). ABC most frequently affects the eccentric metaphyses of long tubular bones, the posterior elements of the vertebral bodies (*11,18*), the pelvis, short tubular bones and less frequently the clavicula, scapula and the tarsal bones (*1,10*). Imaging of choice is the MRI, the CT is useful is cases of compromised stability (*11,13*).

Despite different treatment options like excision, curettage with or without bone grafting, e.g., the clinical course of ABCs is sometimes unpredictable and local recurrences often occur (*4,5,10,13*), while spontaneous regression following incomplete removal is unusual and only incidental (*18*).

Although the reported recurrence rates of ABC imply a potential necessity for routine follow-up, no standardized patient management following primary treatment has been proposed thus far.

The aim of this study was to propose a clinical and radiological follow-up regime based on a review of cases treated at our institutions amended by an extensive review of the available literature with complementary consideration of different treatment options including interventional radiological procedures.

MATERIAL AND METHODS

Twenty-eight cases of primary ABC at different sites were treated and followed up from 2000 to 2013 at two institutions. Secondary and solid ABCs were excluded. Patients were managed by a multidisciplinary team and all included in this retrospective analysis (Table I). The medical records and radiographs were reviewed including demographic data (sex, age at presentation), clinical data (presenting symptom, localisation), radiological data and treatment. Recurrence was defined as a re-appearance of a lesion or a progress of a residual lesion. For lesions that recurred, the date of recurrence(s) and the type of subsequent treatment were noted.

No ethical approval and no special informed consent for participation was required, as evaluated by the ethics committees of the participating hospitals and in accordance with German law, since all data collected are part of daily clinical practice. Research carried out was in compliance with the Helsinki Declaration.

Additionally, a review of the literature was conducted for selected articles with papers dealing mainly with a surgical approach to ABC, including treatment procedure, clinical data and recurrence(s) published from January 1980 to December 2014. This review based on the search terms “aneurysmal bone cyst” and “recurrence” was performed using a full-text electronic journal database (PubMed). The search sets were restricted to humans.

RESULTS

Patient population

Our patient population comprised 16 male and 12 female patients. The average age at diagnosis was 20.6 years (range, 7-59 years). All cases were classified as a primary ABC. They were located in the long tubular bones (16×), in the pelvis (4×), in spine/sacrum (5×), in the talus (2) and in the metatarsal bone (1×).

A slow and gradual onset of pain was found in 18 patients with additionally swelling in two of them. Four patients suffered from pathologic fractures with a sudden onset of pain, in four patients the ABCs were detected by chance, and for four patients the clinical symptoms were not available.

The primary treatment of 22 patients was curettage, with additionally autologous bone grafting in 13 of them, additional instillation of a glycocorticosteroid – now abandoned – in three of them and cement augmentation in one of them. Embolization was used for curative purposes in one case. Follow-up with “watch&wait” after biopsy was done in five patients, who declined (further) surgical therapies.

All patients returned regularly for clinical and radiographic follow-up evaluation (mean 42.2 months ; range : 3-166 months. Overall, six local recurrences (26.1%) were recorded and all

Table I. — Data on 28 patients with primary aneurysmal bone cyst (ABC)

No	Age [y]	Sex	Site	Year of treatment / R	Symptoms	Biopsy	Treatment	Follow- up [m]	Time till R* [m]
1	15	F	Metatarsale	2010	P	Yes	C + A	40	–
2	16	F	Tibia (p)	2006	P	Yes	C + A	100	–
3	18	F	T. ischiadicum	–	P + PF	Yes	W&W	55	–
4	10	M	Os ilium	2006 2007 (R) 2009 (R)	ng ng ng	Yes	C + S C + A C + A	78	31
5	20	F	Acetabulum	2006	P	Yes	C + A	48	–
6	46	M	Os ilium	–	IF	No	W&W	6	–
7	16	F	Femur (d)	2012	P	Yes	C + PMMA	16	–
8	10	M	Humerus (p)	2006	P + PF (5×)	No	W&W	67	–
9	17	M	Tibia (p)	2008	P + Sw	Yes	C + A	54	–
10	11	F	Tibia (p)	2011	IF	Yes	C + A	29	–
11	12	F	Tibia (s)	2011	P + PF	Yes	C	24	13
12	10	M	Femur (p)	2006 2008 (R)	ng	Yes	C + S C + A	72	22
13	15	M	Fibula (p)	2011 2013 (R)	P	Yes	C C	33	30
14	7	M	Spine (L1)	2001 2002 (R)	P	Yes	C + A C + A	132	6
15	59	F	Spine (L3)	2012	P	Yes	C + A	6	–
16	15	M	Humerus (d)	2005 2007 (R)	P + PF (3×)	Yes	C C + S	83	20
17	8	M	Sacrum	2006	ng	Yes	C + S	34	–
18	14	F	Humerus (d)	2004	ng	Yes	C	166	–
19	14	M	Femur (d)	2008	IF	Yes	C + A	19	–
20	21	M	Humerus (p)	2006 2006 (R)	P + Sw	Yes	E E	18	–
21	22	M	Talus	2011	P	Yes	C + A	21	–
22	16	M	Talus	2010 2011(R)	P	Yes	C +A C + PMMA	11	–
23	25	M	Fibula (p)	2012	P	No	W&W	9	–
24	19	M	Femur (d)	2013	P	Yes	C + A	3	–
25	39	M	Spine (L5)	2013	P	No	W&W	7	–
26	25	F	Sacrum	2011	P	Yes	C + A	37	–
27	50	F	Tibia (p)	2011	P	Yes	C+A	3	–
28	37	F	Ulna (p)	2011	IF	No	C	10	–

y = years; F = female; M = male; m = month(s); R = Recurrence; p = proximal; d = distal; s = shaft; T = tuber; P = pain; Sw = Swelling; IF = Incidental finding; ng = not given; PF = pathologic fracture; C = curettage; A = autograft; S = steorid(s); E = Embolization. L = lumbar vetrebra; PMMA = polymethylmethacrylat. W&W = Watch and Wait. * In case of recurrences after revision surgery; the time to recurrence following initial treatment is mentioned (right column).

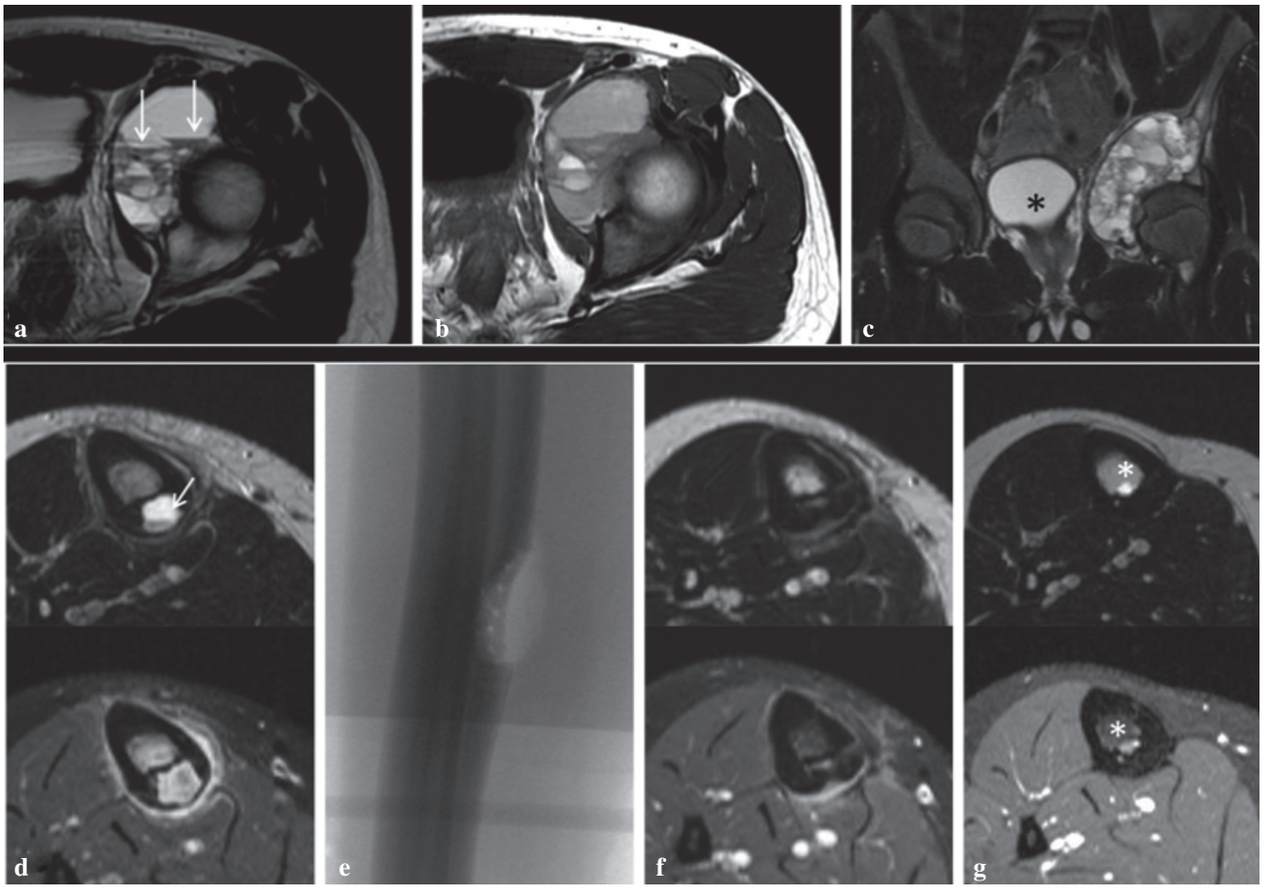

Fig. 1 (a-c). — Typical MR appearance of aneurysmal bone cyst. (a) T2-weighted axial turbo spin echo sequence shows an expansile cystic mass in the left acetabular region with distinct fluid-fluid levels (→). (b) T1-weighted axial turbo spin echo sequence confirms high protein levels within cystic fluid compatible with hemorrhagic changes. (c) Coronal STIR image shows craniocaudal extension of the multiseptated lesion to the hip joint and pelvic cavity with slight displacement of the bladder (*). **(d-g)** Treatment and follow-up of an eccentric, diaphyseal aneurysmal bone cyst (ABC) of the right tibia. (d) Small ABC with typical fluid-fluid-level (→) on axial T2-weighted (top) and contrast-enhanced T1-weighted (bottom) MR images. (e) Intraoperative fluoroscopy image after curettage of the lesion. (f) MR follow-up 3 months after treatment shows only slight enhancement of granulation tissue and confirms complete removal of the ABC. (g) 12 months after treatment, however, a de novo cystic lesion (*) with contrast enhancement can be appreciated at the endosteal margin of the resection site, indicative of recurrent ABC.

of them followed incomplete curettage (mean : 20.3 months (primary recurrence) ; range : 6-31 months). The clinical data are summarized in Table I.

Radiologic hallmarks

In imaging ABCs were expansive and usually eccentrically growing lytic lesions with usually defined margins, marked cortical thinning and no evidence of a solid tumor in the cross sectional imaging. They consisted of blood filled cavernous spaces with internal septation(s) with a high signal intensity on T2 (Fig. 1). After treatment they progressively ossified.

Literature review

Within 1980-2014, we performed a detailed literature analysis via Pubmed of relevant publications, including description of clinical data, surgical approach and recurrence rate, which resulted in

11 papers comprising a total of 790 patients diagnosed with ABC at various sites. These papers are summarized in Table IIa (clinical data) and IIb (treatment and recurrence with (*19*) missing in IIa in reason of absence of clinical data).

Patient age ranged from 1-69 years with most of the patients being diagnosed within the first two decades of life. Localisations mainly comprised the long tubular bones, the spine and the pelvis. Less frequently the short tubular bones, clavicula, scapula, talus and the calcaneus were affected. Clinically, pain was the dominant symptom with an occurrence up to 99%, followed by swelling, pathologic fracture and functional impairment or combination of these. Few ABCs were discovered as an incidental finding. The duration of symptoms was mainly in between 3-6 months, some suffered from pain or swelling < 3 months or up to 24 months. When present a sudden onset of pain was attributable to a pathologic fracture.

Documented treatment was curettage with or without bone grafting (or polymethylmetacrylat, PMMA) in 65.6%, curettage and embolization in 0.25%, en-bloc resection and intralesional curettage in 16.2%, curettage and radiotherapy in 7.7%, radiotherapy in 2.5%, embolization/steroid injection in 2.4% and Amputation in 0.75% patients.

Recurrence rates after curettage varied widely (between 11.8%-59%). The summarized overall recurrences documented in the literature were in patients treated with curettage 17.6%, after curettage and embolization 50%, after en-bloc resection/ intralesional curettage 5.4%, after curettage and radiotherapy 6.6%, 5 after embolization/steroid injection 26.5% and after amputation 0%.

Overall, in the literature documented recurrence occurred between 1-48 months, with the vast majority within the first 2 years. Only few occurred within the third and fourth year after intervention.

DISCUSSION

The ABC is a cystic lesion of bone composed of blood-filled spaces separated by connective tissue septae containing fibroblasts, mononuclear cells, osteoclast-type giant cells and reactive woven bone (Fig. 2a) (*12,14*). It is now widely accepted as a neoplasm owing to recent findings of recurrent chromosomal rearrangements involving the USP6 gene (Fig. 2 b, c) in approximately 70% of cases with restriction to primary ABCs (*20*).

ABCs occur in all age groups, but is most common during the first two decades of life (*8,13,18,22*). Although they can involve any bone of the skeleton, they most frequently affect the long bones, the spine and the pelvis (*1,10,11,18,26*). In imaging ABC is an expansive and eccentrically growing lytic lesion consisting of blood filled cavernous spaces, which progressively ossifies in the reparative phase (*2,19*).

The optimal treatment method selected for an ABC is still being debated (*5*), reflected by the variety of treatment strategies applied in studies published on ABC. Different factors influence the *practical* treatment decision, mainly anatomical site, size of the lesion, and imminence or presence of pathologic fracture (*3,5*). Due to the potentially high destructive capability of the cyst, treatment should be prompt and adequate.

As opposed to intralesional resection techniques wide en-bloc resection has been reported to yield excellent results in terms of local control which approximates 100%, but it may be associated with considerable morbidity and higher complication rates (*5,10,28*). Moreover, wide resection may not always be feasible or unfavourable with regard to the proximity of joints and neurovascular structures, particularly in pelvic, sacrum or vertebra (*5,10*). With regard to the benign nature of the lesion and the existence of less invasive treatment alternatives we propose that wide resection should be restricted to sites in which little functional impairment is to be expected (e.g. fibular shaft) or in cases in which all other treatment options have failed – accepting a higher failure rate concerning recurrence(s). Curettage with bone grafting e.g. is an accepted less invasive surgical treatment strategy. In our study, the recurrence rate after curettage (without any additionally use of radiotherapy and/or embolization, respectively) was 26.1% and in the range of recurrence rates reported by other authors (12%-59%) (Table IIb). One potential factor for recurrence may be attributed to procedures performed using a small bone window with subsequent incomplete removal of the cysts (*7*). Application of high-speed burs has

Table IIa. — Aneurysmal bone cyst (ABC) – clinical data based on literature review

Author (alphabetically)	No.	Age at diagnosis (y)	Localisation	Clinical Symptoms						Duration of symptoms
				P	Sw	P + SW	PF	FI	BC	(m)
Biesecker *et al* (*3*)	66	1. Dec : 39 2. Dec : 50 3. Dec : 13 4. Dec : 3 5. Dec : 1	LTB : 39 Clavicula : 3 STB : 4 Pelvis : 8 Spine : 4 Others : 8	55%	9%	24%	–	8%		< 3 : 33 p 3-12 : 29 p > 12 : 4 p
Boriani *et al* (*4*)	41	5-67 mean : 17.3	Spine : 41	29		11	–	ND : 4 D : 18	–	0.03-2.6 mean 0.8
Cottalorda *et al* (*7*)	21	1.5-5	LTB : 18 Clavicula : 2 Pelvis : 1 Calcaneus : 1	4	1	3	7	2	2	n.g.
Gibbs *et al* (*9*)	40	3-58 mean : 14.5	LTB : 30 Clavicula : 1 Scapula : 2 STB : 3 Pelvis : 4	37	6	–	7	3	1	n.g.
Hay *et al* (*11*)	79	1. Dec : 23 2. Dec : 37 3. Dec : 9 4. Dec : 3 5. Dec : 2 6. Dec : 2 7. Dec : 3	Spine : 79	P; S and Sw in most cases	–	–	–	–	–	14% < 1 30% < 3 mean : 8
De Kleuver *et al* (*12*)	31	7-31 mean : 16	CS : 3 TS : 11 LS : 14 Sacrum : 3	31	8	18	–	S : 19 D : 14 ND : 21	–	
Mankin *et al* (*17*)	150	1-59 mean : 13 Peak in 2 Dec (54.3%)	LTB : 87 Clavicula : 10 Scapula : 3 Spine : 8 Pelvis : 15 Rip : 2 Foot : 8	–	–	–	3	–	–	n.g.
Papagelopoulos *et al* (*21*)	35 (40 in total incl. 5 rec)	3-62	Sacrum 12 Os pubis : 7 Os ischium + Acetablum : 17 Os ilium : 5	38	–	4	7	Pain : + limp : 20 + ROM ↓ : 16 + ND 8/12	–	n.g.
Ruiter et al (*24*)	105	7-31 mean : 18.8	LTB : 51 Spine + sacrum : 17 STB : 15 Flat bone : 22	–	“usually P + Sw”	–	19	–	1	< 3 : 56 p 3-6 : 16 p 6-24 : 15 p n.g. : 18 p
Szendroi *et al* (*25*)	48 (52 in total)	2-44 mean : 15.5 2. Dec : 26	LTB : 33 Scapula : 2 Spine : 4 Hand : 2 Foot : 4	–	–	–	7	–	–	mean : 7
Vergel de Dios *et al* (*27*)	238	1.5-69 mean 16.1 1. Dec : 75 2. Dec : 132 3. Dec : 30 ≥ 5 Dec : 7	LTB and spine : 80% Flat bones : 50% in pelvis Foot : mainly in STB and tarsals (20 p with multiple loc.)	113	43	55	8	–	6	3-48

ABC = Aneurysmal bone cyst; No. = number of patients; p = patients; y = year(s); m = month(s); P = Pain; S = Stiffness; Sw = Swelling; PF = Pathologic fracture; FI = functional impairment; BC = by chance; ND = neurological deficit; D = deformity; LTB = long tubular bones; STB = short tubular bone; CS = cervical Spine; TS = thoracic spine; LS = lumbar spine; n.g. = not given.

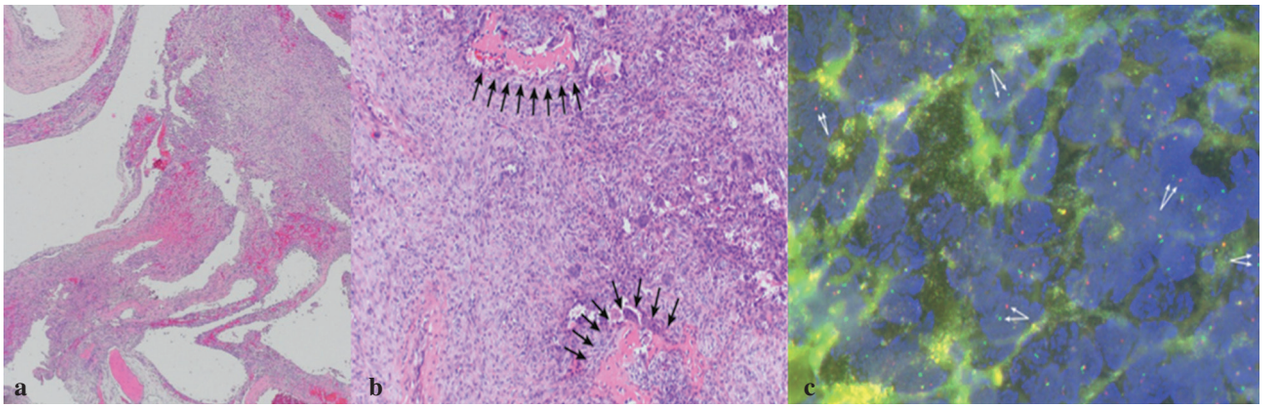

Fig. 2 (a-c). — (a) ABC of the talus showing fibrous septa composed of fibroblast-like mononuclear cells and intermingled multinucleated giant cells. (b) Solid ABC of the proximal femur demonstrating sheets of mononuclear and multinucleated giant cells with varying cellularity. Besides hemorrhage there is marked reactive new bone formation (black arrows). (c) USP6 break-apart FISH shows USP6 rearrangement in the mononuclear cell component (white arrows).

been reported to decrease recurrence rates (*28*). With regard to these studies the authors today favour burring of the bone after curettage through a bone window large enough to visualize the entire cyst whenever possible.

In addition, a variety of non-surgical methods have been proposed for treatment : percutaneous sclerotherapy, embolization, steroid injection, ethibloc® injection, and radiotherapy. These methods may be of particular merit for sites in which surgical therapy is impossible or hazardous (e.g. pelvic, sacral or vertebral ABCs).

Selective arterial embolization (SAE) can be used as an adjunct to surgical treatment to reduce blood-loss during surgery or as stand-alone treatment (*23*). In most cases repeated sessions are required to achieve complete regression without any surgical treatment (*23*). Besides, there are occasional cases in which no arterial feeding vessels suitable for SAE can be identified (*23*). And, although successful treatment with embolization alone has been reported for the pelvis and long bones, its role as the sole mode of therapy in the spine appears to be more limited (*11,23*). Regardless of the number of embolization, 67% of the total patients were considered as cured after a follow-up period longer than 2 years (*23*). All cases of recurrence were within 1 year after the first embolization (*23*), in another study beyond 2 years of embolization (*4*).

Percutaneous sclerotherapy may also be a valuable treatment alternative (*5*). And, administration of polidocanol was found to be a safe and simple procedure with an excellent cure rate and will be especially valuable in ABCs of the pelvis and sacrum (*5*). Like SAE, several sessions are usually required to achieve complete re-calcification of the cyst (*5*). Radiation therapy (20-40 Gy) should not be used because of the risk for post irradiation sarcoma ; rarely it has very limited indications and may remain as an adjuvant therapy for patients with inoperable lesions, aggressive recurrent disease, and medical conditions that place them at a high surgical risk (*10,29*). More recently, denosumab was identified as a potential new and innovative treatment option for ABCs (*15*). However, much longer follow-up and clinical studies are warranted to establish the value of the drug in the treatment of ABCs (*15*).

With regard to follow-up recommendations, it is important to state that 90% of the recurrences occur within 6 to 12 months after initial treatment and rarely after 4 years. With respect to these findings shorter follow-up intervals within the first two years and longer intervals up to the fifth year seem to be reasonable. We therefore propose a routine follow-up regime ideally with MRI and clinical survey performed at 3 (baseline), 6, 12, 18 and 24 months followed by yearly exams in the third to fifth year after

Table IIb. — Aneurysmal bone cyst (ABC)-treatment and recurrence based on literature data

Author (alphabetically)	No. of patients	Treatment and Recurrence(s) [n(treated)/n(recurrence) (%)]						Recurrence(s) In total	Time interval (1. recurrence(s)
		Curettage (+/- bone grafting vs. PMMA)	En bloc resection / intralesional excision	Curettage and Radiotherapy	Radiotherapy	Steroid-injection/ Embolisation / percutaneous sclerotherapy	Amputation		
Biesecker *et al* (*3*)	66	44 / 26 (59%)	8 / 0 (0%)	–	4 / 1 (25%)	7 (Cry) / 1 (14.3%)	1 / 0 (0%)	28 / 66 (42.4%)	Recurrences within 4 y after intervention
Boriani *et al* (*4*)	41	5 / 1 (20%)	13 / 0 (0%)	11 / 0 (0%)	3 / 0 (0%)	4 / 1 (Emb) (25%)		2 / 41 (5%)	1-12 m
Clough *et al* (*6*)	21	15 / 8 (53.3)	3 / 0 (0%)	2 / 0 (0%)	–	–	–	8/ 21 (38.1%)	0-2 y
Cottalorda *et al* (*7*)	21	12 / 2 (16.6%)	3 / 0 (0%)	–	–	2 / 2 (Ster) (100%) 4 / 1 (perSc) (25%)	–	5 / 21 (23.8%)	3-16 m (one patient with multiple rec. with latest 59 m after first intervention)
Gibbs *et al* (*9*)	40	34 / 4 (11.8%)	6 / 0 (0%)	–	–	–	–	4/ 40 (10%)	2-24 m
Hay *et al* (*11*)	79	28 / 7 (25%)	8 / 0 (0%)	34 / 2 (5.8%)	9 / 1 (11.1%)	–	–	10 / 79 (12.7%)	1-12 m (mean 4 m)
Kleuver de *et al* (*12*)	31	25 / 3 (12%) C + Emb: 1 / 1	2 / 2 (100%) (subtotal L)	1 / 0 (0%)	1 / 0 (0%)	1 (Emb) / 0 (0%)	–	6 / 31 (19.6%)	1-21 m
Mankin *et al* (*17*)	150	130 / 29 (22.3%)	20 / 1 (5%)	–	–	–	–	30 / 150 (20%)	1.2 +/- 0.7 y (range; 0.3-3 y)
Papagelopoulos *et al* (*21*)	35 (40 in total including 5 rec)	21 / 5 (23.8%)	14 / 0 (0%)	–	–	–	–	5 / 35 (14.3%)	up to 18 m
Ruiter *et al* (*24*)	105	54 / 28 (51.6%) C + Emb: 1 / 0	13 + 2 (L) / 4 (26.6%)	1 / 0 (0%)	2 / 0 (0%)	1 (Emb) / 0 (0%)	4 / 0 (0%)	32 / 105 (30.5%)	–
Szendroi *et al* (*25*)	48 (52 primary ABC in total)	26 / 7 (26.9%)	22 / 0 (0%)	–	–	–	–	7/ 48 (14.6%)	mean: 14 m
Vergel de Dios *et al* (*27*)	153 own patients (238 in total)	124 / 27 (21.8 %)	16 / 0 (0%)	12 / 2 (16.6%)	1 / 0 (0%)	–	1 / 0 (0%)	29 / 153 (19%)	26 (90%) in between 2 y; 3 later

No = number; C = Curettage; Cry = cryosurgery; RT = Radiotherapy; perSc = percutaneous sclerotherapy; rec = recurrences; p = primary; s = secondary; Emb = Embolization; L = laminectomy; m = month(s); y = year(s).

initial treatment. Using this regime, all cases of recurrence reported in the literature would have been detected irrespective of the presence of symptoms. As individual data are not reported for any of these series the exact interval from recurrence to diagnosis – assuming the proposed regime had been applied – cannot be stated. For our series, however, the application of such a follow-up regime would have detected all cases of recurrence within 6 months or less. Although not specifically addressed in this study the authors advocate a life-long follow-up in cases treated with radiotherapy acknowledging the risk for post irradiation sarcoma.

Appreciation of the strengths and weaknesses of the present study is warranted. First of all one has to acknowledge that ABC is a rare entity. Excluding solid and secondary ABCs further decreased the number of available patients but added to the histologic homogeneity of this series, thus 28 patients were available for this analysis. In this respect, all but four patients who declined to undergo biopsy had histopathological confirmed primary ABC. The meticulous assessment of the available cases with an average follow-up of more than 42 months, amended by a thorough analysis of previously published cases (790 cases) is another strength of the present study. By this means we were able to show the feasibility of the proposed follow-up regimen with regard to timely detection of local recurrence. Nevertheless, the authors emphasize that the proposed follow-up regimen is based on retrospective analyses and further validation utilizing prospectively collected data is warranted.

CONCLUSION

Knowledge of the ABC entity, now widely accepted as a neoplastic lesion, is important, since this lesion is characterized by a high recurrence rate. Despite different treatment options, the risk for recurrence is particularly high within the first 12 months following initial treatment, diminishes over time and is extremely rare after the fourth post-treatment year. With regard to these findings, we recommend that patients with ABC should be continuously followed-up clinically and radiologically until the fifth year following initial treatment and life-long in cases treated with radiotherapy.

List of abbreviations

ABC : Aneurysmal Bone Cyst ; CT : Computed tomography ; MRI : Magnetic resonance imaging ; PMMA : Polymethylmethacrylat ; SAE : Selective arterial embolization.

Conflict of interest

The authors state that there are no financial and personal relationships with other people or organisations that inappropriately influence (bias) the work.

Compliance with Ethical Standards

The use of clinical data was requested and approved by local ethical committee although exams were performed according clinical indications. And, all procedures performed in study were in accordance with the 1964 Helsinki declaration and its later amendments.

Informed consent

All patients gave written inform consent according to the university hospital standard.

REFERENCES

1. **Adler CP.** Aneurysmal bone cyst. In : Adler CP (ed). *Bone diseases*, 3rd ed. Springer, Berlin, 2006, pp 434-439.
2. **Alvas F, James SL, Davies AM, Saiffudin A.** The role of MR imaging in the diagnostic characterisation of appendicular bone tumours and tumourlike conditions. *Eur Radiol* 2007 ; 17 : 2675-2686.
3. **Biesecker JL, Marcove RC, Huvos AG, Miké V.** Aneurysmal bone cysts. A clinicopathologic study of 66 cases. *Cancer* 1970 ; 26 : 615-625.
4. **Boriani S, De Iure F, Campanacci L *et al.*** (2001) Aneurysmal bone cyst of the mobile spine : report on 41 cases. *Spine* 2001 ; 26 : 27-35.
5. **Brosjö O, Pechon P, Hesla A Tsagozis P, Bauer H.** Sclerotherapy with polidocanol for treatment of aneurysmal bone cysts. *Acta Orthop* 2013 ; 84 : 502-505.
6. **Clough JR, Price CH.** Aneurysmal bone cyst : pathogenesis and long term results of treatment. *Clin Orthop Relat Res* 1973 ; 97 : 52-63.
7. **Cottalorda J, Kohler R, Chotel F.** Recurrence of aneurysmal bone cysts in young children : a multicenter study. *J Pediatr Orthop* 2005 ; B-14 : 212-218.
8. **Dahlin DC, McLeod RA.** Aneurysmal bone cyst and other nonneoplastic conditions. *Skeletal Radiol* 1982 ; 8 : 243-250.
9. **Gibbs CP Jr, Hefele MC, Peabody TD *et al.*** Aneurysmal bone cyst of the extremities. Factors related to local recurrence after curettage with a high-speed burr. *J Bone Joint Surg [Am]* 1999 ; 81 : 1671-1678.
10. **Harrop JS, Schmidt MH, Boriani S, Shaffrey CI.** Aggressive “benign” primary spine neoplasms : osteo-

blastoma, aneurysmal bone cyst, and giant cell tumor. *Spine* 2009 ; 34(22 Suppl) : 39-47.

11. **Hay MC, Paterson D, Taylor TK.** Aneurysmal bone cysts of the spine. *J Bone Joint Surg [Br]* 1978 ; 60-B : 406-411.
12. **Kleuver M de, van der Heul RO, Veraart BE.** Aneurysmal bone cyst of the spine : 31 cases and the importance of the surgical approach. *J Pediatr Orthop B* 1998 ; 7 : 286-292.
13. **Kransdorf MJ, Sweet DE.** Aneurysmal bone cyst : concept, controversy, clinical presentation, and imaging. *Am J Roentgenol* 1995 ; 164 : 573-580.
14. **Kyriakos M, Hardy D.** Malignant transformation of aneurysmal bone cyst, with an analysis of the literature. *Cancer* 1991 ; 68 : 1770-1780.
15. **Lange T, Stehling C, Fröhlich B *et al.*** Denosumab : a potential new and innovative treatment option for aneurysmal bone cysts. *Eur Spine J* 2013 ; 22 : 1417-1422.
16. **Leithner A, Windhager R, Lang S *et al.*** Aneurysmal bone cyst. A population based epidemiologic study and literature review. *Clin Orthop Relat Res* 1999 ; 363 : 176-179.
17. **Mankin HJ, Hornicek FJ, Ortiz-Cruz E, Villafuerte J, Gebhardt MC.** Aneurysmal bone cyst : a review of 150 patients. *J Clin Onco*l 2005 ; 23 : 6756-6762.
18. **Nielsen GP, Fletcher JA, Oliveira AM.** Aneurysmal bone cyst. In : In Fletcher CDM, Bridge JA, Hogendoorn PCW, Mertens F (eds) : *WHO Classification of Tumours of Soft Tissue and Bone*. IARC Press, Lyon, 2013, pp 348-349.
19. **O'Donnell P, Saifuddin A.** The prevalence and diagnostic significance of fluid-fluid levels in focal lesions of bone. *Skeletal Radiol* 2004 ; 33 : 330-336.
20. **Oliveira AM, Chou MM.** USP6-induced neoplasms : the biologic spectrum of aneurysmal bone cyst and nodular fasciitis. *Hum Pathol* 2014 ; 45 : 1-11.
21. **Papagelopoulos PJ, Choudhury SN, Frassica FJ *et al.*** Treatment of aneurysmal bone cysts of the pelvis and sacrum. *J Bone Joint Surg [Am]* 2001 ; 83-A : 1674-1681.
22. **Rosenberg AE, Nielsen GP, Fletcher JA.** Tumours of undefined neoplastic nature. In : Fletcher CDM, Unni KK, Mertens F (eds). *Pathology and Genetics of Tumours of Soft Tissue and Bone*. IARC Press, Lyon, 2002, pp 338-339.
23. **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 : 161-167.
24. **Ruiter DJ, van Rijssel TG, van der Velde EA.** Aneurysmal bone cysts : a clinicopathological study of 105 cases. *Cancer* 1977 ; 39 : 2231-2239.
25. **Szendröi M, Cser I, Kónya A, Rényi-Vámos A.** Aneurysmal bone cyst. A review of 52 primary and 16 secondary cases. *Arch Orthop Trauma Surg* 1992 ; 111 : 318-322.
26. **Tillman BP, Dahlin DC, Lipscomb PR, Stewart JR.** Aneurysmal bone cyst : an analysis of ninety-five cases. *Mayo Clin Proc* 1968 ; 43 : 478-495.
27. **Vergel De Dios AM, Bond JR, Shives TC, McLeod RA, Unni KK.** Aneurysmal bone cyst. A clinicopathologic study of 238 cases. *Cancer* 1992 ; 69 : 2921-2931.
28. **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 : 1649-1659.
29. **Wiklund TA, Blomqvist CP, Räty J *et al.*** Postirradiation sarcoma. Analysis of a nationwide cancer registry material. *Cancer* 1991 ; 68 : 524-531.