Current treatments of primary aneurysmal bone cysts

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The management of aneurysmal bone cyst depends on the age of the patient, the location, extent, aggressiveness and the size of the lesion. In the light of their experience and a review of the literature of 1256 aneurysmal bone cysts, the authors analyzed various treatment modalities. Inactive lesions can heal with biopsy or curettage alone. In active or aggressive lesions, elective treatment usually consists of curettage, whether associated or not with bone grafting and local adjuvants. Aneurysmal bone cyst in young children do not seem more aggressive than in older children. In pelvic locations, the emergence of a few cases of spontaneous healing (even in active or aggressive lesions) encourages the adoption of clinical and radiological supervision for some months after biopsy when possible. In some cases, the localization and extent of the cyst are such that operative treatment is extremely hazardous. Selective arterial embolization has made a considerable contribution towards the therapeutic solution of such cases. For some authors, direct percutaneous Ethibloc injection can be recommended as the first-choice treatment except in spinal lesions. Nevertheless, the complications encountered in some series after percutaneous embolization of aneurysmal bone cyst with Ethibloc should encourage the use of Ethibloc injection not as an initial treatment but as a reliable alternative to surgery. *J Pediatr Orthop B* 15:155–167 © 2006 Lippincott Williams & Wilkins.

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Introduction

Aneurysmal bone cyst (ABC) is a benign cystic lesion of bone composed of blood-filled spaces separated by connective tissue septa containing fibroblasts, osteoclast-type giant cells and reactive woven bone [1]. Jaffe and Lichtenstein [2] established ABC as a distinct clinical entity in 1942 by clearly separating it from hemangiomas of the bone and from other tumors in which giant cells were also a prominent feature. ABC can exist either as a primary bone lesion (in about 70% of the cases), or as a secondary lesion when a pre-existing osseous lesion can be identified (in about 30% of the cases) [3]. ABC represents approximately 1% of all bone tumors [3]. Primary ABC is a rare lesion with an incidence of 0.14 per 10^5 individuals [4]. Although ABC may be observed at any age, it distinctly predominates from 10 to 20 years of age. Approximately three-quarters of all cases are observed under 20 years of age. Primary ABC rarely occurs after 30 years of age, and it is exceptional after 50 years [5]. In demographic data on 411 children with primary ABCs, the femur (22%), tibia (17%), spine (15%), humerus (10%), pelvis (9%) and fibula (9%) were the most common locations [6].

The early radiographic appearance of ABC is a subperiosteal, poorly defined osteolysis, elevating and inflating the periosteum and progressively eroding the cortex [3]. Magnetic resonance imaging (MRI) is helpful in showing the fluid–fluid levels within the cyst and the internal septation of the cyst, with soap-bubble features (particularly in T2-weighted images) with contrast enhancement of the septa [3].

The usual methods of treatment are curettage [7–17], resection [7,8,10–12], intracystic injections [18–28] and embolization [29–36]. Opinion varies considerably as to which method should be used, particularly in children. Most of the larger series reported in the literature include both children and adults [7,9–12]. Some exclusively pediatric studies exist, but all of these reported less than 50 patients [37–46]. The biggest are those of Dormans *et al.* [37] (45 children) and Ramirez and Stanton [38] (29 children with follow-up). Given that individual centers did not have an adequate volume of cases, we performed a multihospital retrospective study of 156 primary ABCs in children and adolescents [6]. In the light of our experience [3,6,40,45,47–49] and a review of the literature (1256 primary ABCs), we analyzed various treatment modalities. Even if our experience only concerns pediatric fields, the different methods of treatment of primary ABCs are quite the same in adults and in children. Moreover, the treatment of primary ABC in adults is often less difficult than in children because of the absence of the growth cartilage and of the lower recurrence rate [9].

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Biopsy

Even if plain radiographs and MRI often support the diagnosis of ABCs, accurate histologic evaluation with correlation of radiographic and MRI findings is imperative for definitive diagnosis. In typical cases in which no impairment is envisioned, some surgeons opt for the 'one-stage' approach without carrying out a biopsy a priori. Nevertheless, we believe a suspected ABC should not be treated without first performing a biopsy [3,48]. Tomeno *et al.* [50] reported that out of 51 cases of ABCs, the initial diagnosis based on radiological analysis in 17 cases was that of a unicameral bone cyst. Puncture of the cyst is not sufficient to make the diagnosis. Clear liquid is often observed in solitary bone cysts but it can also be found in an inactive ABC [5,9,11,39,45]. Conversely, bloody liquid can be observed in an ABC but also in fractured solitary bone cysts [39,45]. In Bonakdarpour's series [51] concerning primary and secondary ABCs, five independent observers reviewed 26 radiographs of secondary ABC. The consensus was that in five cases the first diagnostic choice was ABC, despite the histologic findings of another lesion in each case. As ABCs can arise in or mimic a malignancy (namely telangiectasic osteogenic sarcoma), a biopsy is imperative before treatment. Imaging diagnosis remains a diagnosis of probability and should always be confirmed by a biopsy [39,45,52].

Various forms of therapy

Abandoned or little-used treatments

Radiation therapy

Radiation therapy proved to be effective in inducing cyst ossification but it carries the risk of growth arrest, subsequent limb-length inequality and radiation-induced sarcoma [5,15,53,54]. This procedure must now be avoided because of such side effects.

Kirschner pins

The Papavasiliou and Sferopoulos [55] approach of inserting Kirschner pins protruding beyond the cortex and the periosteum prevents the creation of a closed bone cavity, which is believed to be the main cause of recurrence following other types of treatment. Even though his study was based on only six children, the results were promising. The authors concluded that it is worthwhile considering the insertion of Kirschner pins as a first step in the treatment. To the authors' knowledge, there are no other studies involving more cases. Nevertheless, if breaking up the existing equilibrium in the blood-filled cavities by perforation of the bone wall with Kirschner pins could explain the promising results, it is not likely that the pins really prevent the creation of a closed cavity. As a matter of fact, pins will rapidly be coated by ossification and the cavity will quickly be closed.

Demineralized bone particles

For Delloye and co-workers [25,56] ABC can be considered a reparative process with cells in the septa having an intrinsic potential for new-bone formation. Delloye and co-workers [25–56] thought that the introduction of an osteoinductive material into the cyst might trigger the ossification and hence the healing of the cyst. They used demineralized bone and autogenous bone marrow as an inducer of the healing phase with the expectation that the cells of the ABC septa would be responding cells. One of the advantages of such a method is that curettage is unnecessary, thereby allowing for a minimally invasive approach. Delloye and co-workers [25–56] reported 13 cases of induced healing of ABCs following this method of treatment. Successful healing was observed in 11 cases after a mean duration of follow-up of 3.9 years. As no curettage is required, the proposed treatment avoids extensive surgery and blood loss and is convenient for the treatment of poorly accessible lesions such as those occurring in the pelvis. The results of this treatment are promising.

Methylprednisolone acetate injection

Treatment by perforation of the bone wall with cannulated needles introducing methylprednisolone acetate into the cyst was reported with negative results [39,48,57]. Scaglietti *et al.* [57] tested corticosteroid injections in the treatment of 12 ABCs. In each patient, at least three successive injections were performed in a period of 8 months. The results were negative for the 12 patients. The lesion can show signs of further development after corticosteroid injections [39,48,57,58]. Steroid injection is not indicated in patients with ABC [48].

Percutaneous alcoholic injection

Glorion *et al.* [20] reported seven cases of ABCs treated with intracystic injections of pure alcoholic solution (not diluted) with 2 years follow-up. In six cases the cysts healed. This treatment is currently under evaluation but initial results are promising.

Percutaneous calcitonin injection

The role of calcitonin in the treatment of ABC is still hypothetical; it may block the activity of the osteoclasts and/or promote the formation of new bony trabecula in the fibrous septa of the ABC. For Szendroi *et al.* [23] most ABCs are shown to be hypovascular by angiography and are therefore unsuitable for superselective embolization. In these cases, calcitonin hormone therapy administered directly into the cyst results in progressive ossification and healing of the ABC [23]. Six out of seven hypovascular ABCs responded well to the calcitonin administered directly into the cyst but the maximum follow-up was only 14 months [23]. Three ABCs healed completely and another three incompletely. In one case, the hormone therapy had to be interrupted because of allergy. Calcitonin was administered three times a week

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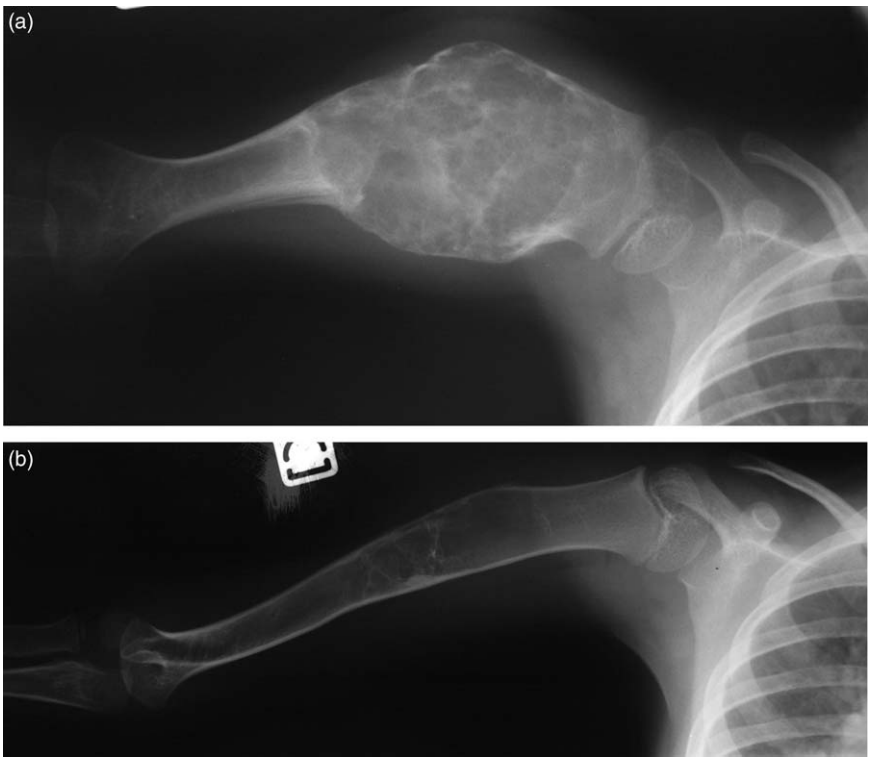
(15 times altogether) by direct puncture of the cyst under local anesthesia. Calcitonin can be administered under local anesthesia because of the thinned and softened bony wall of the cysts [23].

Embolic methods

Percutaneous embolization with an alcoholic solution of zein

Alcoholic zein (Ethibloc, Ethnor Laboratories/Ethicon, Norderstedt, Germany) is a radiopaque alcoholic solution of corn protein. Ethibloc is a fibrosing and sclerosing agent, probably without any specific osteogenic property [59]. It is a biodegradable agent that induces intravascular thrombosis, marked local inflammation, and finally a fibrotic reaction [18,21]. The aim is to bring a sclerosing substance in contact with the cystic wall to obstruct at the venous side the multiple parietal arteriolar afferents of the ABC [26]. Percutaneous embolization is performed as follows [21]: with the patient under general anesthesia, a needle is inserted with fluoroscopic or computed tomographic (CT) guidance. Cyst fluid is aspirated and contrast medium is injected into the lesion to confirm good filling of the cyst, absence of leakage in the surrounding soft tissue and to show venous drainage. Assessment of venous drainage is less accurate with CT guidance than with fluoroscopic control. If only partial filling is achieved, additional injections are performed to ensure complete filling. Reassessment of the venous drainage is mandatory after each additional injection. Potential venous drainage is controlled with compression (manual or with a tourniquet) to prevent distal embolization of the venous draining system, and then alcoholic zein is injected. The ensuing reparative process, which results in bone reconstruction (Fig. 1) and bone volume normalization, lasts several months and even over 1 year after percutaneous injection. Sometimes, several injections are necessary [19,21]. This treatment remains highly controversial. For some authors [18,21] Ethibloc injection is an efficient and safe treatment in primary ABCs and can be considered a reliable alternative to surgery, especially in cases with a difficult surgical approach or in cases of postsurgical recurrence. For Garg *et al.* [19], Falappa *et al.* [26] and Sales de Gauzy *et al.* [60] direct percutaneous Ethibloc injection can be recommended as the first-choice treatment except in spinal lesions. Nevertheless, the complications encountered in some series after percutaneous embolization of ABC with Ethibloc encourage some authors to use Ethibloc injection not as an initial treatment but an alternative to surgery. Ethibloc injection, however, should only be performed by a medical team with a significant experience of this treatment.

Fig. 1



(a) Initial plain radiograph shows an expansile, lucent lesion in contact with the growth plate with numerous septa in the upper end of the humerus in a young child (collection Pr. G. Lefort). (b) Plain radiograph after percutaneous embolization with alcoholic zein without recurrence at more than 5 years follow-up but with residual geodes.

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Table 1 Complications and results of Ethibloc treatment

Authors	Number of cases	Transitory local inflammatory reactions with local pain	Pulmonary embolism	Aseptic osteitis	Aseptic abscess or cutaneous fistulae	Several injections[a]
Adamsbaum *et al.* [18]	17	16	0	0	3	3 (up to 3)
Garg *et al.* [19]	10	2	0	0	1	3 (up to 2)
Guibaud *et al.* [21]	16	5	1	1	0	6 (up to 3)
Falappa *et al.* [26]	13	8	0	0	2	9 (up to 4)
Topouchian *et al.* [24]	15	5	1	0	4	4 (up to 3)
Sales de Gauzy *et al.* [60]	12	5	0	0	0	1 (up to 2)
Total	83	41	2	1	10	26 (up to 4)

Authors	Total improvement	Partial improvement	No bone reconstruction or secondary surgery
Adamsbaum *et al.* [18]	14	0	3
Garg *et al.* [19]	7	3	0
Guibaud *et al.* [21]	13	1	2
Falappa *et al.* [26]	13	0	0
Topouchian *et al.* [24]	9	2	4
Sales de Gauzy *et al.* [60]	6	3	3
Total	62	9	12

[a]The first number is the number of patients who needed supplementary injections and the number in brackets is the maximal number of Ethibloc injections.

The complications and results of the major series are summarized in Table 1. Transitory local inflammatory reactions with fever and local pain limited to the injection site are common [18]. It is more a side effect than a complication. It is important to protect the skin so as to prevent a leakage of alcoholic zein into soft tissue resulting in local cellulitis. Falappa *et al.* [26] believe that careful asepsis, preliminary injection of hydrosoluble contrast medium, simultaneous monitoring during Ethibloc injection, 10 min of manual pressure on the injection site and postprocedural antibiotic therapy could prevent major complications.

Topouchian *et al.* [24] reported five major complications in 15 children using direct Ethibloc injection. One patient had a pulmonary embolism, which resolved within 7 days in the intensive care unit. Four patients experienced cutaneous fistulization with discharge of the fibrosing agent; all four patients underwent repeated surgery with abscess and fibrosing agent excision. Three out of the four patients with fistulization had cysts located superficially, which led the authors to abandon this technique initially in superficial lesions. The fourth patient, who had a deep lesion location, however, also experienced fistulization and was still experiencing recurrences at 3 years follow-up despite multiple surgical procedures. This led the authors to definitively abandon this technique in the treatment of ABCs even for deep lesion location. Mascard and Adamsbaum [61] was surprised by this article because of the high rate of local and general complications, with these data contrasting with previous reports. Mascard and Adamsbaum [61] noted that this article raised some questions about the technical conditions of Ethibloc injection. The published images of Ethibloc infiltration in soft tissues surrounding bone seem to indicate that excessive amounts of Ethibloc were used, leading to the reported complications. Pulmonary embolism is a serious complication described in three papers [21,24,49]. Another major complication has been described in the literature. An Ethibloc injection in ABC of C2 was performed in a 4-year-old boy [22]. Subsequent CT and MR images demonstrated embolization material in the vertebrobasilar system. The child died of brain stem and cerebellar infarction 23 h after the intervention. Venous drainage being the main risk of Ethibloc injection, the injection must not be used in dangerous locations.

Selective arterial embolization

The aim of embolization is occlusion of the vascular supply to the lesion without interfering with the vascularity of surrounding tissues or structures [29]. Selective arterial embolization (SAE) has been reported as effective treatment and can be considered in lesions whose site (spine, pelvis) or size makes other types of treatment difficult or hazardous [32–36]. This is particularly helpful at sites where it is not possible to use a tourniquet [31]. Successful embolization of an ABC will result in progressive ossification within 2–4 months of initial embolization, which almost always begins peripherally. SAE has several advantages over other forms of treatment. First, it is a nonsurgical technique that may be effective as primary treatment but, in the event of failure, does not preclude the possibility of intervening surgically. Complete ossification may require 8–12 months or longer [29]. SAE may cause complications, but when performed by an experienced interventional radiologist with appropriate precautions these can usually be avoided [31]. Ischemia to neural structures and vital structures, such as the viscera, is the main concern [31]. Neurologic

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complications have been reported to occur when common vessels supplying the vertebra and spinal cord are occluded. For this reason, some authors recommend performing somatosensory-evoked potentials with a provocative amobarbital test before embolization of spinal lesions [30]. Boriani *et al.* [62] advises against the use of SAE in thoracic locations because of the risk of thoracic cord ischemia after the Adamkiewicz artery.

Curettage or saucerization

According to Vergel de Dios *et al.* [7], the most appropriate treatment is curettage with or without bone grafting. In a review of the literature of some series (Tables 2 and 3), the recurrence rate of the 690 ABCs treated with curettage ± bone graft was 31%. In Tillman's series [9] (95 cases), all recurrences except one developed within 2 years of treatment, suggesting that radiographic surveillance for local recurrence should continue for at least 2 years. The mitotic index of the initial lesion did not correlate with that of the recurrent lesion [42]. Recurrence rates after curettage vary widely. It is important to differentiate curettage with a small hole in the cyst wall [41], curettage through a large cortical window that allows excellent observation of the interior of the cyst [37] (usually 70–90% of the diameter of the involved bone) and saucerization [43] to understand the significant variation in the recurrence rate in the literature. The saucerization procedure entails excising the new subperiosteal bone with its attached cyst contents and curettage of the remaining cortical bone. The new subperiosteal bone is removed to allow unrestricted access for curettage of the surface of the remaining normal strut of cortical bone. The cavity is then bone grafted, and the periosteum is closed. Saucerization is recommended when there is a normal strut of cortical bone to maintain the overall length and alignment of the long bone. [41,43] In Cole's study [41], curettage was performed through a small hole in the cyst wall. As a result, the contents of the cyst might have been incompletely removed, and this would account for the high failure rate of this procedure. In contrast, saucerization provided direct access to the whole cyst and enabled complete removal of the soft tissue contents [41]. All the ABCs treated with this procedure healed [41]. Saucerization and subperiosteal resection are almost identical procedures, as the cyst is only surrounded by a thin incomplete layer of subperiosteal new bone [41]. This review of the literature shows that curettage through a small opening must be avoided. One must operate with a proper fenestration/saucerization, large enough to enable the surgeon to control the procedure. Otsuka *et al.* [63] used endoscopic curettage in the treatment of ABC. For them [63], endoscopic curettage requires only two small incisions. The cosmetic results are excellent. Moreover, the thinned bone cortex is minimally disturbed, which preserves the bone strength. New bone formation is facilitated by the minimal damage to the bone and the soft tissue. Close-up endoscopic imaging also allows for more thorough curettage. Whether the graft is autogenous or allogenous makes no significant difference in the recurrence rate [9].

On account of the high risk of recurrence after curettage alone, several authors used various forms of adjunctive therapy, such as phenol, polymethylmethacrylate, liquid nitrogen or the use of a high-speed mechanical burr to decrease the rate of local recurrence. In Tables 2 and 3, the lower recurrence rate with adjunctive therapies shows the efficiency of these treatments.

- Embolization can be used as a treatment alone or before surgical procedure to reduce the vascularity of the lesion, thus facilitating a more complete excision [29–31,34]. This is particularly helpful in sites where it

Table 2 Different types of initial treatment and recurrence rates of aneurysmal bone cyst (ABC) in general series

Authors	Number of ABC	Curettage ± grafting	Curettage and cementation	Cryotherapy ± curettage	Curettage + radiation therapy	Curettage with a high-speed burr	Radiation therapy alone	Marginal or wide resection
Campanacci *et al.* [12]	161	91 (19)	–	–	15 (3)	–	8 (2)	47 (0)
Vergel de Dios *et al.* [7]	153	124 (27)	–	–	12 (2)	–	1 (0)	16 (0)
Mankin *et al.* [17]	150	110 (24)	20 (5)	–	–	–	–	20 (1)
Marcove *et al.* [54]	106	44 (26)	–	51 (9)	–	–	11 (1)	–
Ruiter *et al.* [11]	105	82 (28)	–	–	–	–	2 (0)	21 (4)
Osaki *et al.* [67]	65	30 (11)	35 (6)	–	–	–	–	–
Biesecker *et al.* [10]	63	44 (26)	–	7 (1)	–	–	4 (1)	8 (0)
Gibbs *et al.* [68]	40	–	–	–	–	34 (4)	–	6 (0)
Nobler *et al.* [15]	33	18 (6)	–	–	1 (0)	–	6 (1)	8 (2)
Schreuder *et al.* [52]	27	–	–	27 (1)	–	–	–	–
Clough and Price [69]	21	15 (6)	–	–	2 (0)	–	1 (0)	3 (0)
Koskinen *et al.* [14]	20	14 (2)	–	–	1 (0)	–	–	5 (0)
Server Perez *et al.* [86]	20	17 (4)	–	–	1 (0)	–	–	2 (0)
Farsetti *et al.* [16]	20	11 (2)	–	–	3 (0)	–	–	6 (0)
Total	984	600 (181)	55 (11)	85 (11)	35 (5)	34 (4)	33 (5)	142 (7)[a]

The first number is the number of cases and the number in parentheses is the number of recurrences. The series by Tillmanns *et al.* [9] and Capanna *et al.* [13] are not in this table because they are earlier reports on the same patient population as the series by Vergel de Dios *et al.* [7] and Campanacci *et al.* series [12], respectively.
[a]All the recurrences were in marginal resections and not for wide ones.

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Table 3 Different types of initial treatment and recurrence rates of aneurysmal bone cyst in exclusively pediatric series

	Number of cases	Curettage ± grafting	Saucerization alone	Saucerization + high-speed burr + grafting +/ adjuvant therapies	Curettage and cementation
Dormans *et al.* [37]	45	–	–	44 (8)	–
Ramirez and Stanton [38]	29	24 (8)	–	–	–
Arlet *et al.* [39]	28	11 (4)	–	–	–
Bollini *et al.* [40]	27	12 (5)	–	–	–
Cole [41]	25	18 (7)	4 (0)	–	–
Epeldegui Torre [44]	14	13 (4)	–	–	–
Ozaki *et al.* [46]	14	5 (1)	–	–	5 (1)
Freiberg *et al.* [42]	7	7 (5)	–	–	–
Total	189	90 (34)	4 (0)	44 (8)	5 (1)

	Curettage + radiation therapy	Radiation therapy alone	Ethibloc	Resection	Other treatment
Dormans *et al.* [37]	–	–	–	1 (0)	–
Ramirez and Stanton[38]	–	–	–	5 (0)	–
Arlet *et al.* [39]	–	2 (0)	–	14 (0)	1 (healing without treatment)
Bollini *et al.* [40]	1 (0)	–	3 (1)	11 (0)	–
Cole [41]	1 (0)	–	–	2 (0)	–
Epeldegui Torre [44]	–	–	–	–	1 (healing after injection of prednisolone)
Ozaki *et al.* [46]	–	–	–	4 (0)	–
Freiberg *et al.* [42]	–	–	–	–	–
Total	2 (0)	2 (0)	3 (1)	37 (0)	2

The first number is the number of cases and the number in parentheses is the number of recurrences. The series by Cottalorda *et al.* [45] is not in the table because it is an earlier report on the same patient population as the series by Bollini *et al.* [40].

is not possible to use a tourniquet to control bleeding [31]. SAE is the most practical tool to use exclusively, or in an initial phase, or in preparation for surgery.

- Phenol is a nonselective cytotoxic agent, and when applied directly to the surface of curetted tumors it destroys the remaining residual tumor and normal cells [64,65]. Recurrence is probably due to its superficial action and the impossibility of penetrating the periphery beyond the surgical margin [52].
- Cryotherapy consists of local intralesional excision followed by application of liquid nitrogen [54]. After curettage, liquid nitrogen is poured into the cavity, taking care to protect the skin to prevent necrosis. Marcove *et al.* [54] treated 51 ABCs with curettage and cryotherapy. They reported nine local recurrences. The overall cure rate was 96% after a second, or repeat, cryosurgery. Schreuder *et al.* [52] treated 27 ABCs with curettage and cryotherapy. They reported only one local recurrence. The advantage, compared with local resection, is that the supportive function of bone is preserved and reconstructive surgery can be limited [52]. Complications reported with this treatment include superficial skin necrosis, deep wound infection, fractures, flap necrosis, mild joint stiffness, osteonecrosis, injury to adjacent neurovascular structures, gas embolism and growth arrest [52,54,66]. These complications can be limited with a good technique.
- The rationale for the use of polymethylmethacrylate cement as adjuvant treatment is based on its heating effect [67]. Experiments have shown that a thermal lesion of at least 50° is necessary for a cytotoxic effect [52]. The maximum peripheral extent of a thermal lesion varies from 2.5 mm in cancellous to 0.5 mm in cortical bone [52]. One advantage of cementation compared with other methods is that bone cement supports the mechanical stress involved and prevents fracture [67]. In addition, there is an important point in follow-up after cementation. If a bone graft is performed immediately after curettage, radiologic detection of local relapses become very difficult. The cement provides the advantage of early radiographic evidence of local recurrence. In cementation, follow-up with MRI scans can also reveal local recurrences more clearly than after grafting. If a local relapse is identified, curettage and cementation can be performed at an early stage [67]. Destruction of the growth plate or subchondral bone by thermal injury can be prevented by using artificial skin patches or bone graft [67]. As most patients with ABCs are children or young people, Ozaki *et al.* [67] think that the packed cement must be removed. In addition, there is the advantage that a second-look biopsy can be performed during removal of the cement.
- Gibbs *et al.* [68] described curettage with the use of a mechanical burr in 34 patients having an ABC of an extremity. They reported four local recurrences. They concluded that rates of local control of almost 90% can be achieved with this association in patients who have an ABC of an extremity. According to most orthopaedic oncologists, curettage (hand curette) and burr drilling are no more two separate procedures because both are performed subsequently.

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Resection

En-bloc resection is the treatment with the lowest risk of local recurrence [7,10,38,69] (Tables 2 and 3): seven recurrences for 179 resections. All the recurrences were in marginal resections and not in wide ones. The local relapse rate after wide resection was 0% in the series by Campanacci *et al*. [12] (47 resections) and Vergel de Dios *et al*. [7] (16 resections). Resection is not a 'usual' procedure, but must be considered only when there is no other alternative. En-bloc resection can be the treatment of choice in eccentric lesions or in lesions arising in nonessential, expendable bones (proximal fibula, clavicle, rib, pubic ramus) [12]. In other locations, this treatment is accompanied by loss of bone and the need for reconstruction with associated morbidity [52]. In a benign condition that is amenable to more conservative methods, even though less dependable its use can be justified only when resection will not significantly interfere with function [69]. It is occasionally advisable to resort to reconstruction using a vascularized fibula bone graft [49]. In children, subperiosteal resection is possible [40,45]. The remaining periosteum will over several months contribute to good bone consolidation. In adults or, alternatively, in certain very bulky lesions, it is not technically viable to perform subperiosteal resection because the periosteum cannot be isolated.

Specific locations and problems

Pelvic location

In demographic data on 411 children with ABC, the pelvis was involved in 36 cases (8.8%) [6]. ABC of the pelvis is a challenging therapeutic problem [70]. Special factors need to be considered in the management of ABCs of the pelvis; these include the relative inaccessibility of the lesions, associated intraoperative bleeding, the proximity of the lesions to neurovascular structures (excision of a pelvic ABC may be difficult when the lesion extends around iliac vessels, the lumbar plexus or the sciatic nerve) and the vulnerability of the integrity of the acetabulum. Healing following biopsy of two ABCs of the pelvis in two boys aged 10 and 11 years has been described [71]. In the study by Capanna *et al*. [72], two patients, aged 13 and 15 years, had only incisional biopsies, as their parents refused further treatment. Although both lesions appeared radiographically aggressive, with expansion into the soft tissues, the disease did not progress. On the contrary, the lesions became quiescent with reconstitution of a radiographic bony shell. The same evolution has been noted by several authors [41,47,73]. Complete curettage is probably not necessary, and simple removal of the soft tissue contents is sufficient to induce healing [41]. It appears that, in some way, breaking up the existing equilibrium in the blood-filled cavities (e.g. simple biopsy or incomplete curettage) may be sufficient to cause involution of the lesion. In most cases, when a pelvic ABC is diagnosed a curative treatment is rapidly performed. Had the two patients in the series by Capanna *et al*. [72] been operated on, it would not have been possible to observe spontaneous healing. It could be possible that spontaneous healing is not quite as rare as described in the literature (four out of 15 cases in the series by Cottalorda *et al*. [47]) because most of them are treated rapidly after diagnosis. The emergence of a few cases of spontaneous healing (even in active or aggressive lesions) encourages the promotion of clinical and radiological supervision during some months after biopsy when possible. In the event of propitious evolution, supervision must be continued and surgery may be avoided. If the lesion increases, treatment must be proposed. The reasons for the apparent difference in the behavior of long bone and pelvic cysts have not been determined [41]. In the series by Papagelopoulos *et al*. [70], 40 consecutive patients with an ABC of the pelvis and/or sacrum were treated from 1921 to 1996. Complete intralesional excision, including exteriorization of the lesion and removal of the overlying cortex of the entire lesion so that the cavity can be seen clearly, was performed in a systematic, thorough manner [70]. Acetabular lesions were associated with such complications as pathological fracture, protusio acetabuli and hip dislocation [70]. Perhaps one explanation for these major complications lies in less safe and effective treatment in older cases. The patients might also have been seen and treated at a later and more evoluted stage. In the study by Capanna *et al*. [72], significant complications affected the final functional result in four out of the seven patients who received radiation therapy (older cases), while only one minor complication was seen in the surgical group.

Substantial intraoperative bleeding is common [70]. Embolization can be used as a treatment alone or before surgical procedure to reduce the vascularity of the lesion, thus facilitating a more complete excision [29–31,70]. This is particularly helpful in pelvic locations [31].

Spinal location

Lesions with spinal involvement often pose very difficult problems in treatment, especially when presenting with cord compression [74]. The first is related to biopsy. In lesions with a major hemorrhagic component, as with ABC, needle biopsy can be misleading and an open biopsy is therefore suggested [62]. In these cases, curettage should be performed at the time the biopsy is carried out [62,75] For Papagelopoulos *et al*. [76], 12 out of the 40 patients with a primary spinal ABC were diagnosed incorrectly from the initial needle biopsy specimen.

Adjuvant therapy, such as preoperative embolization, is often necessary. Excessive intraoperative blood loss can be substantial and one patient died despite the resuscitation efforts in the series by Papagelopoulos *et al*. [76]. As ABC often affects the pediatric spine

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(63% in the series by Boriani *et al.* [62]), aggressive surgery for this benign, sometimes self-repairing lesion should be avoided [62]. For Boriani *et al.* [62], SAE alone is the first treatment option for spinal ABC because it can be as effective as intralesional surgery, although minimally invasive, and is the treatment with the best cost-to-benefit ratio. They did not use SAE in thoracic locations because the risk of thoracic cord ischemia after Adamkiewicz artery embolization is controversial [62]. Embolization as sole treatment may address the spinal lesion, but it cannot address a concomitant deformity or an instability.

In case of neurologic involvement, pathologic fracture, technical impossibility of performing embolization or local recurrence after at least two embolization procedures, surgery represents the ideal therapy [62]. Most authors recommend curettage, which represents a less invasive surgery [62,75–78]. The satisfactory results obtained after complete intralesional excision of primary vertebral lesions indicate that wide resection by total spondylectomy is not necessary for these lesions [76].

Several authors [77,79] reported the devastating effects of treating the tumor in isolation without addressing the concomitant deformity and instability. If instability or deformity or both are present, instrumentation and fusion should be performed at the time of the surgical resection or before other forms of therapy (Fig. 2). When fusion is needed, titanium instrumentation must be used and is recommended to allow high-quality MRI surveillance of the region for signs of recurrence after surgery [80,81]. Spinal fusion with instrumentation is strongly recommended if more than two facets (more than one on each side, or more than two full facets) are violated during the excision of ABC [80]. Despite the risk of postoperative deformity, in children, several authors [62,75] prefered to use an orthesis until skeletal maturity before performing an arthrodesis.

A case of a wide excision of a thoracic ABC using a cavitron has been described [82]. This device was especially helpful in the achievement of an extensive excision of a relatively inaccessible lesion of the spine.

The surgical approach depends on the location of the cyst. Posterior approaches provided insufficient access to lesions located in the vertebral body and were associated with a higher recurrence rate than anterior approaches [79]. Two surgical approaches (anterior and posterior) are often necessary because the lesions are expansive. In case of neurologic involvement, the treatment consists, of a laminectomy first and secondarily an anterior approach, even if the compression is anterior [79].

Local recurrences in the spine (which occur in 10–25% of cases), particularly in aggressive lesions, are unpredictable and may have grave consequences [62,76,80,83,84]. Garg *et al.* [80] (in 12 children) noticed that a surgical technique using a four-step approach of intralesional curettage, high-speed bur, electrocautery and bone grafting had a significantly reduced rate of recurrence (0/8 cases) compared with that of traditional intralesional curettage and bone grafting (4/4 cases). Short-segment spinal fusion with instrumentation performed at the time of initial surgery was associated with a decreased risk of local recurrence (0/5 cases) versus those without a spinal fusion at initial surgery (4/7 cases). While not a statistically significant finding, it showed a strong trend toward significance ($P < 0.08$). This is probably not a direct causal relationship but may instead be related to the wide surgical exposure that allowed excellent visualization for excision of the ABC at the expense of requiring a short-segment spinal fusion [80].

Lesions in contact with growth cartilage

Lesions in contact with the growth cartilage have the potential to create serious complications in treatment [38,40]. The major complications are physeal arrests [38,40]. It is difficult to say whether the growth plate lesion was due to the cyst itself or caused by the surgical procedure instead [16]. Ramirez and Stanton [38], using MRI, noted that ABC frequently crosses the physis before any surgical procedure (Fig. 3). Some of the postoperative growth arrests noted in surgical series before the availability of MRI evaluation might not have been iatrogenic in nature [38]. Even though physeal arrest can occur after treatment, juxtaphyseal ABC can be treated satisfactorily with intralesional surgery and bone grafting with the expectation of normal physeal growth [85].

A reluctance on the part of the surgeon to perform a sufficiently extensive procedure in or near an open growth plate, especially in the lower extremity, could result in residual tissue after curettage, thus facilitating local recurrence [68]. Either the curettage is wide and the risk of epiphysiodesis is considerable or the curettage is limited and the risk of recurrence is high [45]. A careful curettage must be performed at a distance from the growth plate and then with a blunt dissector when in contact with the growth plate in order not to damage it [45]. Even if some surgeons prefer to prevent local recurrence rather than lose the epiphyseal plate, we do not agree with this approach. In children, and especially in young ones, the cartilage must be preserved at all costs, including the risk of having a recurrence because of insufficient curettage [45]. These children will be kept under supervision. It is easier to treat a possible recurrence recognized at an early stage than an epiphysiodesis.

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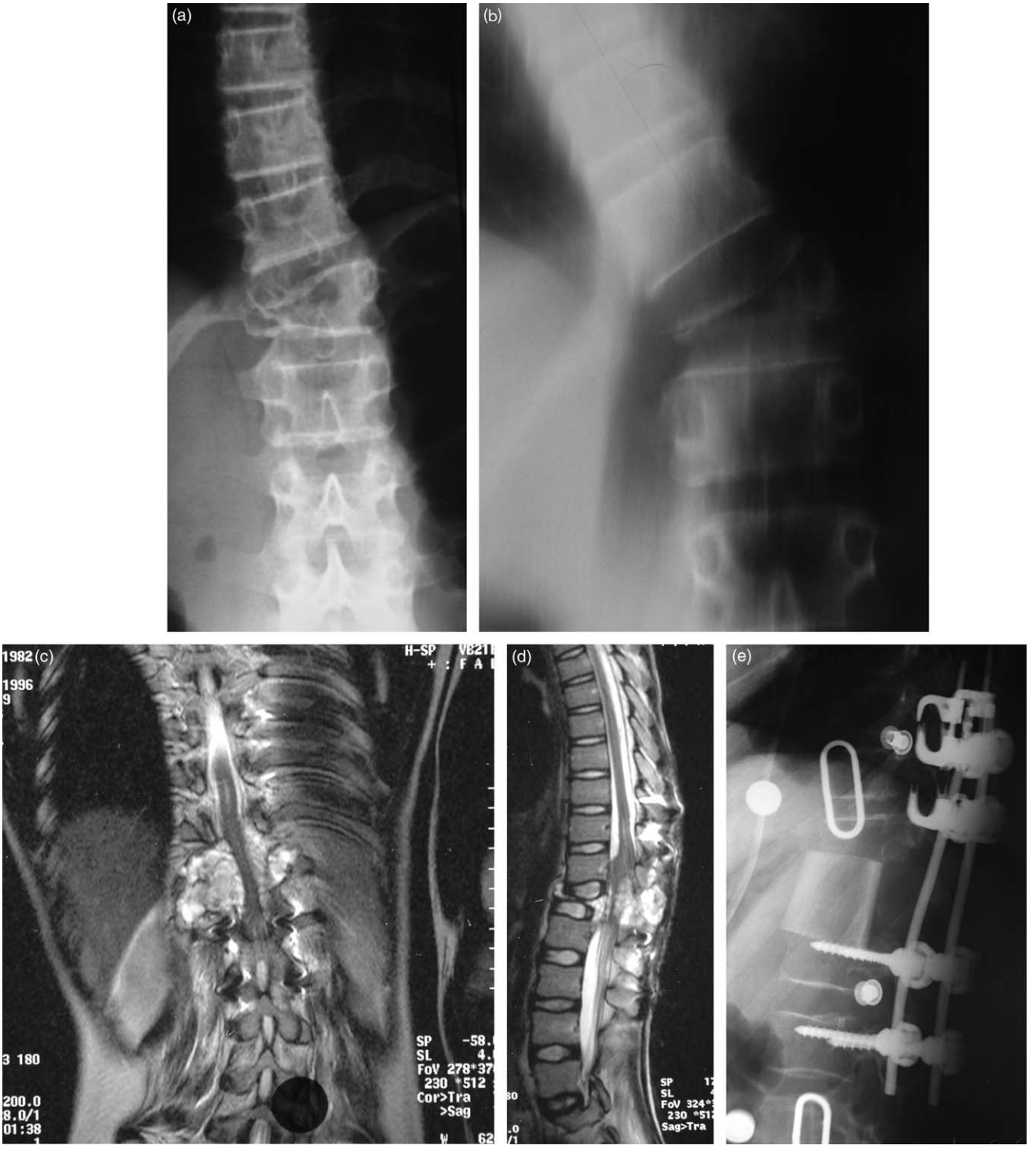
Fig. 2



(a) Aneurysmal bone cyst involving T12. This child presented with a painful scoliosis with a paraplegia (collection Pr. G. Lefort). (b) The tomography shows this vertebral collapse. (c) Anteroposterior preoperative magnetic resonance imaging (MRI). (d) Lateral preoperative MRI. (e) Lateral postoperative radiograph after surgical resection with tibial anterior bone graft and posterior arthrodesis. Four months later, a total neurologic recovery was observed.

Younger patients

The patient's age is reported to be one of the risk factors for local recurrence [7,9,42]. Is it necessary to adapt the treatment to the age of the patient? Several authors reported that recurrences occur more often in younger patients [7,9,10,38,42]. In the study by Vergel de Dios

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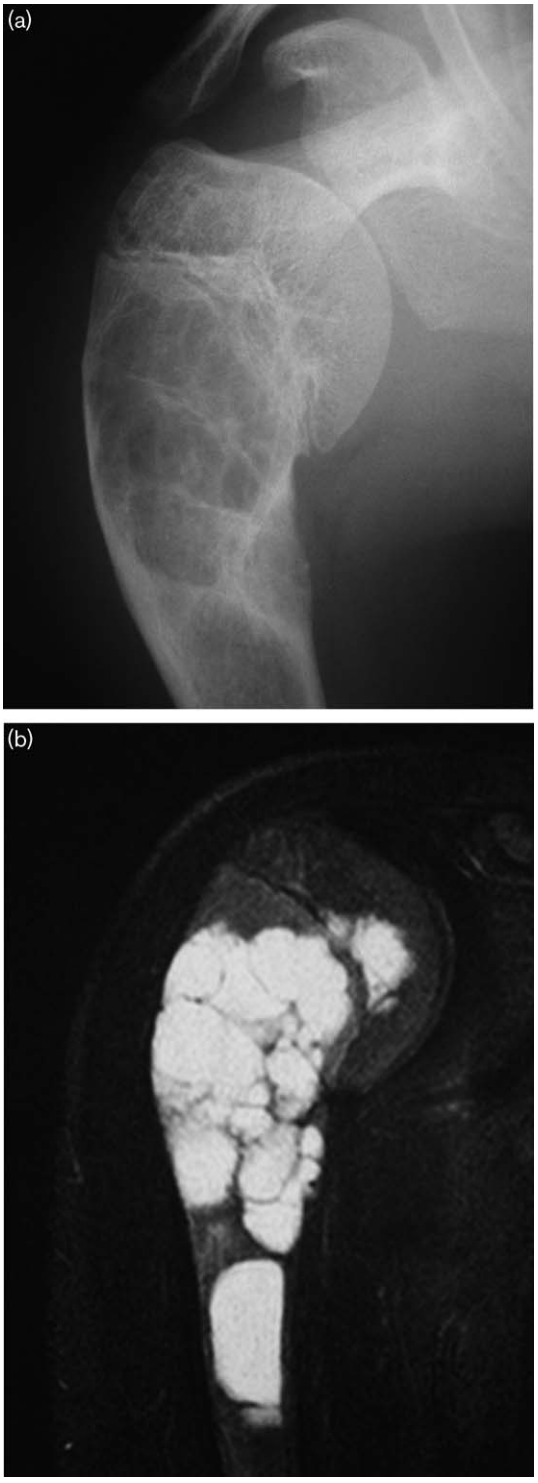
et al. [7] (238 ABC), more than 93% of recurrent lesions occurred in patients under 20 years of age. Tilmann *et al*. [9] (95 ABC), in an earlier report on this same patient population, observed that the risk of recurrence diminished with advancing age (32% recurrences under 15 years, 12% after 15 years and 0% after 25 years). For Biesecker *et al*. [10] (66 ABC), there is a higher probability of recurrence in younger patients (≤ 15 years). All these series noted differences in recurrence rates between children (≤ 15 years) and adults. Gibbs *et al*. [68] noted a significantly higher rate of recurrence in patients with open growth plates, suggesting that a young age and open physes are indicators of a poor prognosis relative to local recurrence. Is there a definitive difference in recurrence rate between young children and older ones? The highest recurrence rate (three children out of four) in Ramirez and Stanton's series [38] was in children under 5 years of age (four children out of 29). According to Ramirez and Stanton, [38] one explanation for this finding is that the clinical and pathologic behavior of ABC in younger patients is more aggressive. Another interpretation of the data suggests that the higher recurrence rate is due to excessive caution on the part of the surgeon when faced with a potential growth arrest in such a very young patient. Freiberg *et al*. [42] in their series (seven children under 10 years of age) noted five recurrences after initial curettage and bone grafting; this rate was 100% in the aggressive and active lesions (five cases). Although only concerned with very few cases, these results induced some authors to have a more 'aggressive' surgical attitude in younger children. Studies on a larger scale did not confirm those results. Ozaki *et al*. [46] recorded three recurrences in their series of 14 children under 10 years of age (but only one under 5 years). Dormans *et al*. [37] studied among 45 children whether recurrence rates were higher in younger patients. The children were studied and classified according to their age group (≤ 10 years of age or > 10 years of age). The difference in persistence or recurrence rates on age was not statistically significant. Cottalorda *et al*. [48] (21 children, aged 5 years or less) found only two recurrences out of 14 curettages. In fact, the clinical and pathologic behavior of ABC in younger patients does not seem to be more aggressive than that in older children. More aggressive treatment does not, therefore, seem to be justified.

Fig. 3



(a) Anteroposterior radiograph of an aneurysmal bone cyst (ABC) crossing the growth cartilage with epiphysiodesis. (b) magnetic resonance imaging of the same ABC.

Residual geodes

Sometimes, residual geodes cause concern about possible recurrence, but after periods of observation the final state of these children can be considered close to recovery (healing manifested by increased cortical and septal thickening) (Fig. 1b) [47]. It is therefore important to avoid excessively early repeat surgical procedure to treat a possible recurrence when faced with a residual geode. Several authors [26,39,47,54] also noticed that ABC often healed with small static residual cysts. Evaluation of residual geodes based on plain radiographs certainly underestimates geodes because they are mostly not seen on plain radiographs but only with CT imaging [47].

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Treatment options

Capanna *et al.* [13] described three radiographic grades. Inactive cysts have a complete periosteal shell with defined sclerotic bone limits. Active cysts have an incomplete periosteal shell and defined bone limits. Aggressive cysts have an indefinite margin and show uniform osteolysis. All the recurrences in their series were in active or aggressive tumors. No recurrence was observed in the inactive types. This is important for the treatment.

As described in this review, the methods of treatment in the literature vary widely and treatment of an ABC remains highly controversial. We believe that a suspected ABC should not be treated without biopsy [47]. The management of ABC depends on the age of the patient, the location, the vascularity of the lesion, the size of the lesion and how far it has extended. Each case is unique and as such there is no 'uniform' recipe. In the light of the literature, however, a certain degree of consensus is emerging regarding the following propositions:

- Inactive lesions can simply be monitored without any treatment [3,5]. Sometimes, these lesions heal without recurrence with just biopsy or curettage [3,5]. Consequently, it is not necessary to propose a more 'aggressive' treatment initially and the children will be kept under supervision [47,70]. In the event of propitious evolution, supervision must be continued and surgery may be avoided. If the lesion increases or if it is painful, treatment must be proposed.
- For active or aggressive lesions, the surgical indication will depend on tumor location [86]. The rate of recurrence after curettage alone being high, resection offers a satisfactory theoretical solution. Marginal en-bloc resection may be adopted in eccentric lesion or in expendable bones (fibula, rib, pubic ramus) with little morbidity and minimal risk of recurrence [7,10,38]. Exceptionally, the expansion on the lesion will require the resection of an important osteo-articular segment. The position of the cyst, however, limits the indications: it is not always easy in long bones, if the tumor is juxtaposed to the growth plate, difficult in the pelvis when it is not situated in the ischium and often impossible in the spine [45]. Consequently, the most common elective treatment consists of curettage, associated or not with bone grafting. Of course, there is always a risk of recurrence, but the recurrence rate can be reduced with local adjuvants. For some authors, ABC between 10 and 20 years of age (the majority of the cases) corresponds to the giant cell tumors of more than 20 years of age, in almost all aspects. The treatment approach with respect to pathology is identical, that is, it is imperative to sterilize the cavity walls to prevent local recurrence.
- In some cases, the localization and extent of the cyst are such that operative treatment is extremely hazardous. Resection is sometimes impossible and curettage can be extremely difficult with considerable blood loss. SAE, in isolation or before surgery, or Ethibloc injection makes a considerable contribution to the therapeutic solution of such cases [18,19,21, 29–31]. With regard to the complications in some series after percutaneous embolization of ABCs with alcoholic zein [24], Ethibloc injection must not be considered as an initial treatment but as an alternative to surgery, especially in cases with a difficult surgical approach or in cases of postsurgical recurrence.
- Other treatments remain of marginal use. It is hard to introduce major changes in the standard of care without prospective studies (but prospective studies are quite impossible because of the incidence of ABC) and hard data to suggest that an alternative technique is truly superior.

Conclusion

As primary ABC often affects children, aggressive surgery for this benign, sometimes self-repairing, lesion should be initially avoided. On the other hand, its benign nature should not occult its local aggressivity, which may cause neurologic complications, damage to the growth plate and therapeutic problems associated with the frequently large size of these ABCs. Therefore, it is essential that after initial treatment these patients be kept under supervision for several years to detect a recurrence.

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