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The symptomatology of aneurysmal bone cyst. The value of diagnostic imaging

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Summary

Background: An aneurysmal bone cyst (ABC) is a benign lesion occurring mainly in children. Generally it is found in the metaphyses of long bones and in flat bones. Osteolysis, bone expansion, thinning of the cortex, osseous septa and fluid levels belong to the typical imaging findings in ABC cases. The purpose of the study was to determine the value of imaging methods: plain x-ray films, US, CT and MRI for the diagnosis of aneurysmal bone cyst and to work out the diagnostic algorithm for an ABC suspected on plain radiograms.

Material/Methods: The material consists of 72 patients (44 boys, 28 girls) aged 2–23 years (mean age 12 yrs) with bone lesions diagnosed as aneurysmal bone cysts on pathologic examination. In all patients, plain radiographs were performed, in 26 – sonography (US), in 41 – computed tomography (CT), in 8 – magnetic resonance imaging (MRI). All the examinations were analyzed according to own system of evaluation of typical presentation in the particular methods. The histopathological diagnosis was established after biopsy or surgery.

Results: The aneurysmal bone cysts were most frequently located in the long bones of the lower limbs (43%), in the long bones of the upper limbs (26.4%) and in flat bones (25%). The localization was mainly in the bone methaphyses (56.6%).

On pathological examination, primary ABC was diagnosed in 65.3% of cases, secondary ABC in 23.6%, ABC and concomitant osteitis in 7%, ABC and giant cell tumor in 2.7%, ABC and chondrosarcoma in 1.4%.

The application of four methods resulted in 72.2% consistency between diagnostic imaging and pathological examination. In case of only one method used the consistency was 75% for MRI, 63% for CT, 55.4% for plain films and 50% for US. In 100% of misdiagnosed cases there were no septa within osteolytic lesions on plain films, whereas in 43.8% features of malignancy were found. In 53% of misdiagnosed cases no fluid levels were observed on CT and in 69% on US.

Conclusions: 1. The diagnosis of aneurismal bone cyst based on plain radiography is possible in cases with typical radiological signs. 2. Application of other methods significantly increases the percentage of correct diagnoses. 3. Lack of fluid levels does not exclude the diagnosis of ABC. 4. Imaging features of malignancy do not exclude the diagnosis of ABC.

Key words: aneurysmal bone cyst (ABC) • X-rays, ultrasound (US) • computed tomography (CT) • magnetic resonance imaging (MRI)

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Background

An aneurysmal bone cyst (ABC, cystis aneurismatica), according to WHO classification a tumor-like lesion, is rare and accounts for 1-6% of primary bone tumors [1–3]. 80–90% of such lesions appear below 20 years of age, usually in the second decade of life [1,2,4,5]. They can present as primary tumors developing in previously unaffected bone, or secondary lesions, preceded by other processes both benign and malignant [3,6]. ABC was first described by Jaffe and Lichtenstein in 1942 [1,4], however, the etiopathogenesis of the lesions is still unclear [2,4]. Local hemodynamic disturbances such as venous occlusion or arteriovenous shunts, associated with previous traumas, developmental anomalies, tumors or inflammatory processes are believed to play a role in the development of ABC [2,7–9]. It may develop in various locations of the skeletal system [1,9–15], most frequently in long bone metaphyses, in flat bones and posterior elements of the vertebral bodies [2,3,16]. The clinical presentation depends on the location of the lesion. They include pain, bone expansion, increased temperature and local sensitivity, as well as impaired mobility. In case of lesions located in the vertebral column, neurological signs can be observed. In the bone, an ABC is usually located eccentrically, on the borderline between the cortical layer and the marrow cavity, or subperiosteally. It consists of blood-filled cysts, communicating with each other, separated by highly vascularized septa with fibrous structure, containing polynuclear giant cells, osteoblastic and mature osseous tissue. Focal or scattered hemosiderin deposits can also be observed within the septa [2,4]. Aneurysmal bone cysts are characterized by rapid growth. They can reach considerable sizes, with large periosseous component, especially in flat bones and in the vertebral column. Differential diagnosis should include giant cell tumors, isolated cysts, fibrous dysplasia, chondromyxomas, osteoblastomas, chondromas, sarcomas, predominantly osteosarcomas and lesions associated with hyperparathyroidism and hemophilia [2,3]. The basic method of treatment of ABC is surgery, with curettage or resection of the lesion, often with subsequent site reconstruction with a bone implant. Other methods of treatment include selective embolization of blood vessels and injections of substances stimulating fibrotic processes and blood clotting into the tumor mass [1–3,5,17]. The prognosis in ABC is good, although some patients (10–30%) develop relapses, which are the most common problem encountered in the course of treatment [5,17]. Postoperative recurrences are usually observed during the first two years after the surgery [1]. Sporadically, malignant transformation of the process at various interval after treatment may occur; therefore, long-term clinical observation or control imaging is recommended [2,5]. Despite different imaging modalities used, ABC may sometimes cause diagnostic problems.

Aim of the study

The purpose of the study was to determine the value of imaging methods: plain radiography (x-ray), ultrasound (US), computed tomography (CT) and magnetic resonance imaging (MRI) in the diagnosis of aneurysmal bone cyst and to work out the diagnostic algorithm for an ABC suspected on the basis of plain radiograms.

Table 1. Histopathological diagnosis.

Histopathological diagnosis	Number of patients	Percentage
Primary ABC	47	65.3%
Secondary ABC (due to fibrous dysplasia, non-osseous fibroma, chondroblastoma, chondroma, trauma)	17	23.6%
ABC + osteitis	5	7.0%
ABC + giant cell tumor	2	2.7%
ABC + chondrosarcoma	1	1.4%

Table 2. Localization of the ABC.

ABC location	Number of patients	Percentage
Long bones, lower extremities	31	43.0%
Long bones, upper extremities	19	26.4%
Flat bones	18	25.0%
Vertebral body, metacarpals	1+3	5.6%

Table 3. Localization of the ABC within the bone.

ABC location in the bone	Number of patients	Percentage
Metaphysis	30	56.6%
Diaphysis	15	28.3%
Metaphysis and diaphysis	4	7.6%
Metaphysis and epiphysis	2	3.8%
Epiphysis, metaphysis and diaphysis	1	1.8%
Epiphysis	1	1.8%

Material and Methods

The material consisted of 72 patients referred to the Institute of Mother and Child in 2000–2007 with tumor-like bone lesions, who were diagnosed with aneurysmal bone cysts on the basis of histopathology, The group comprised 44 boys and 28 girls aged 2–23 years. The average age was 12 years and patients between 10 and 19 years of age constituted the most numerous age group (67%).

In the diagnostic period, all the patients underwent plain radiography in two projections, 26 – sonography

Table 4. Imaging features of ABC on plain radiographs, US, CT and MRI.

Symptoms	Plain radiography (n=72)		CT (n=41)		US (n=26)		MRI (n=8)	
Bone expansion	62	(86.0%)	25	(61.0%)	–		3	(37.5%)
No expansion	10	(14.0%)	16	(39.0%)	–		5	(62.5%)
Single osteolytic focus	47	(65.3%)	25	(61.0%)	9	(35.0%)	6	(75.0%)
Numerous cyst-like lesions	25	(34.7%)	16	(39.0%)	17	(65.0%)	2	(25.0%)
Septa	32	(44.5%)	14	(34.0%)	–		3	(37.5%)
No septa	40	(55.5%)	27	(66.0%)	–		5	(62.5%)
Cortex thinning	43	(60.0%)	12	(29.2%)	–		3	(37.5%)
Cortex disruption	6	(8.3%)	20	(48.8%)	–		3	(37.5%)
Cortex thinning & disruption	13	(18.0%)	6	(7.5%)	–		1	(12.4%)
Cortex unaffected	10	(13.8%)	3	(7.3%)	–		1	(12.4%)
Periosteal reaction	26	(36.0%)	15	(36.5%)	5	(19.2%)	3	(37.5%)
Malignant periosteal reaction	4	(5.6%)	4	(10.0%)	9	(34.6%)	0	
No periosteal reactions	42	(58.4%)	22	(53.5%)	12	(46.1%)	5	(62.5%)
Signs of malignancy*	15	(21.0%)	7	(17.0%)	9	(34.6%)	2	(25.0%)
No signs of malignancy	57	(79.0%)	34	(83%)	17	(65.3%)	6	(75.0%)
Fluid levels	–		11	(26.8%)	9	(35.0%)	4	(50.0%)
No fluid levels	–		30	(73.2%)	17	(65.0%)	4	(50.0%)
Soft tissue tumor	10	(13.8%)	7	(17.0%)	6	(23.0%)	2	(25.0%)
Soft tissue thickening	–		7	(17.0%)	5	(19.0%)	2	(25.0%)
No lesions	–		27	(66.0%)	15	(58.0%)	4	(50.0%)
PD – significantly increased flow	–		–		3	(11.5%)	–	
PD – moderate flow	–		–		6	(23.0%)	–	
No flow	–		–		10	(38.5%)	–	
Not investigated	–		–		7	(27.0%)	–	
Atypical contrast enhancement	–		7	(17.0%)	–		2	(25.0%)
Signal in images T1 ↓, T2 ↑	–		–		–		8	(100.0%)

* Obscured borderlines of the osteolytic lesion, disrupted cortical layer with ill-delineated periosseous mass, disrupted periosteum, Codman's triangle, pathological contrast enhancement.

Table 5. Consistency of imaging and histopathological diagnoses.

Imaging modality	Correct diagnoses/ number of examinations	Percentage rate
All imaging modalities	52/72	72.2%
Plain radiography	40/72	55.5%
CT	26/41	63.0%
US	13/26	50.0%
MRI	6/8	75.0%

(US), 41 – computed tomography (CT), 8 – magnetic resonance imaging (MRI). All the examinations were analyzed according to own system of evaluation of typical presentation in the particular methods, developed in the Department of Diagnostics Imaging of the Institute of Mother and Child. The anatomical localization of the lesion was also taken into consideration. The consistency between the provisional diagnoses based on imaging studies and the histopathological diagnoses was analyzed. The histopathological diagnoses were established in the Department of Pathomorphology of the Institute of Mother and Child on the basis of biopsy or surgical specimens.

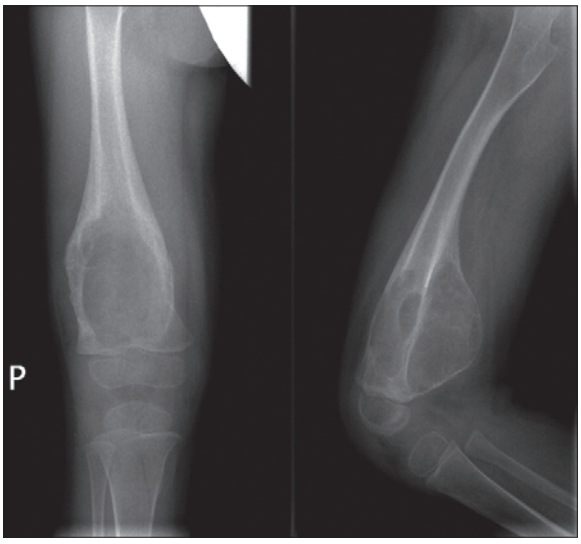


Figure 1. Typical appearance of an ABC on plain radiograph; trauma in anamnesis.

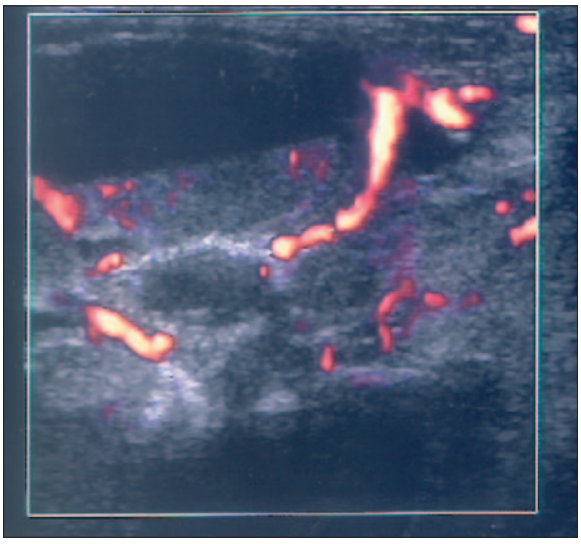


Figure 2. Typical sonographic appearance of ABC; fluid levels, vascularity of the septa.

Results

Table 1 presents histogical diagnoses. Localization of ABC in the skeletal system is presented in Table 2. Localization in the bones is illustrated in Table 3. Table 4 presents imaging characteristics of ABC in the particular methods. Consistency of diagnoses based on imaging modalities and histopathology is illustrated in Table 5.

Analysis of misdiagnosed cases

On plain radiograms, among 32 patients with results inconsistent with histopathology results, in 32 cases (100%) no septa were observed, in 25 (78%) a single osteolytic focus and in 14 (43.8%) signs of malignancy such as obscured borderlines of the osteolytic lesion, disrupted cortical layer with an ill-delineated periosseous mass, periosteal disruption and Codman's triangle. Out of 15 CT-based misdiagnoses, in 8 cases (53.3%) no septa and in 8 no fluid levels were visible, in 7 (46.7%) signs of malignancy were observed (cortex disruption, ill-delineated periosseous tumor, periosteal disruption, Codman's triangle, contrast enhancement). Among 13 cases misdiagnosed on the basis of US scans, in 9 (69%) no fluid levels were observed, in 6 (46%) there were signs of malignancy (cortical layer disruption with soft tissue tumor, periosteal disruption, enhanced flow in PD). In two cases, one localized in the calcaneal bone and the other in the vertebral body, ABC was not diagnosed by MRI. In both cases a single osteolytic focus without septa and fluid levels, with inhomogeneous contrast enhancement and a tumor-like lesion in soft tissue was observed – both these cases were suggestive of a malignant lesion.

Among 20 cases misdiagnosed on the basis of imaging in 9 malignancy was suspected, in 6 a cyst-like lesion suggestive of a simple bone cyst, in 2 an osteolytic focus of unclear character, in 2 osteitis, and in 1 a chondroma.

Conventional radiography is the first and basic examination in the diagnostics of ABC. Ultrasound should be performed more often, because of the capability of visualizing cyst-like areas, fluid levels, and excluding the signs of malignancy. Their availability, non-invasive character and short duration are also important advantages. CT and MRI, because of their properties, generally allow to establish the diagnosis in dubious cases. They are also indicated for the determination of tumor size, relation to the adjacent anatomic structures, exclusion of fractures and other complications.

Discussion

Many manuals and publications describe the typical presentation of an ABC in imaging modalities [1–3,7,15]. Plain radiograms show a polycyclic osteolytic lesion with balloon-like bone expansion, often with spaces divided by osseous septa, located asymmetrically, eccentrically, or peripherally in long bones, well-delineated, with sclerotic layer on the periphery (Figure 1). Periosteal reactions are usually absent, except for regenerative ones after fractures [1,7]. An ABC causing thinning and disruption of the cortical layer can be assessed easily by US, which can visualize morphology of the process, tissue elements and cyst-like spaces with fluid levels (Figure 2), which, according to many authors, are characteristic and present in most cases [6,9,14]. Fluid levels develop as a result of separation of cellular elements from blood serum filling the cysts. Their presence is confirmed by CT and MRI, which also visualize osseous septa [1,6,7,16]. However, fluid levels are not pathognomonic for ABC, they can be observed also in malignant processes. Both MRI and CT can usually exclude malignant character of the lesion [2,4,16]. CT enables to assess the character of lesion in the cortical layer of the bone and periosteum, the sclerotic layer on the periphery of soft tissue pseudotumor, representing calcified to various extent, detached periosteum, is visible [2,16]; in flat bones, ABC tends more frequently to expand outside the bone [7]. Generally no significant contrast enhancement is observed in CT. In MRI, ABC shows decreased or intermediate signal intensity in T1-weighted images, which can, however, become high in case of severe bleeding. In T2-weighted images the signal intensity is high or average, with inhomogeneous hyperintense

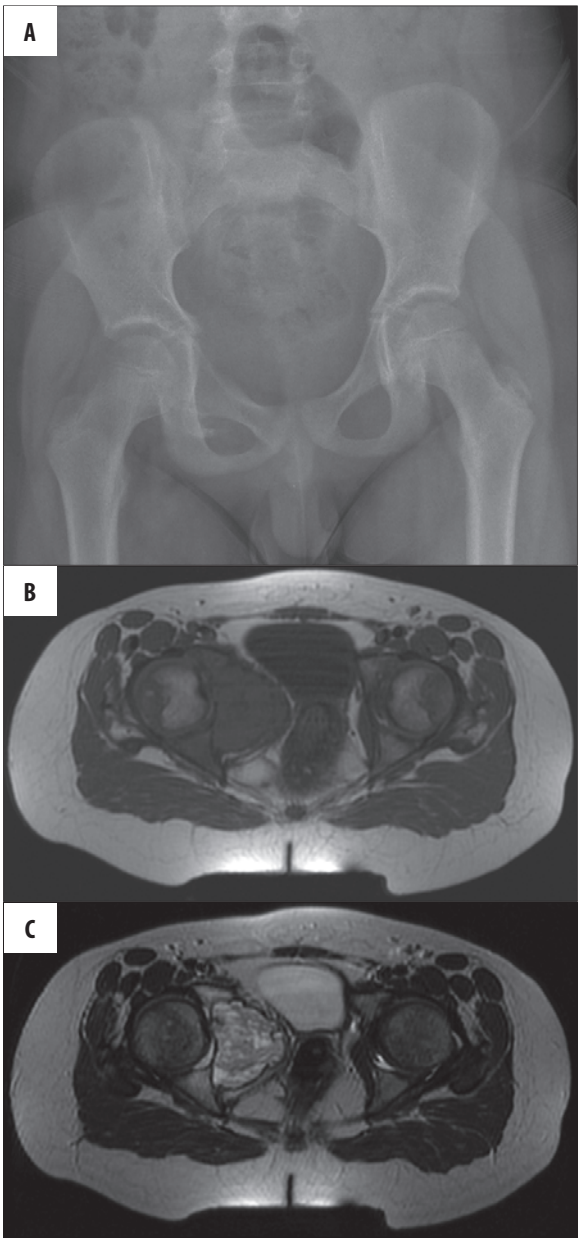


Figure 3. Patient B.J. (**A**) Plain film. Suspected malignant tumor of the right pubic bone. Ill-defined mass around the pubic bone, Codman's triangle. (**B,C**) MRI. ABC of the pubic bone: no signs of malignancy, well-defined mass, hypointense on T1WI (**A**), inhomogenously hyperintense on T2WI (**B**).

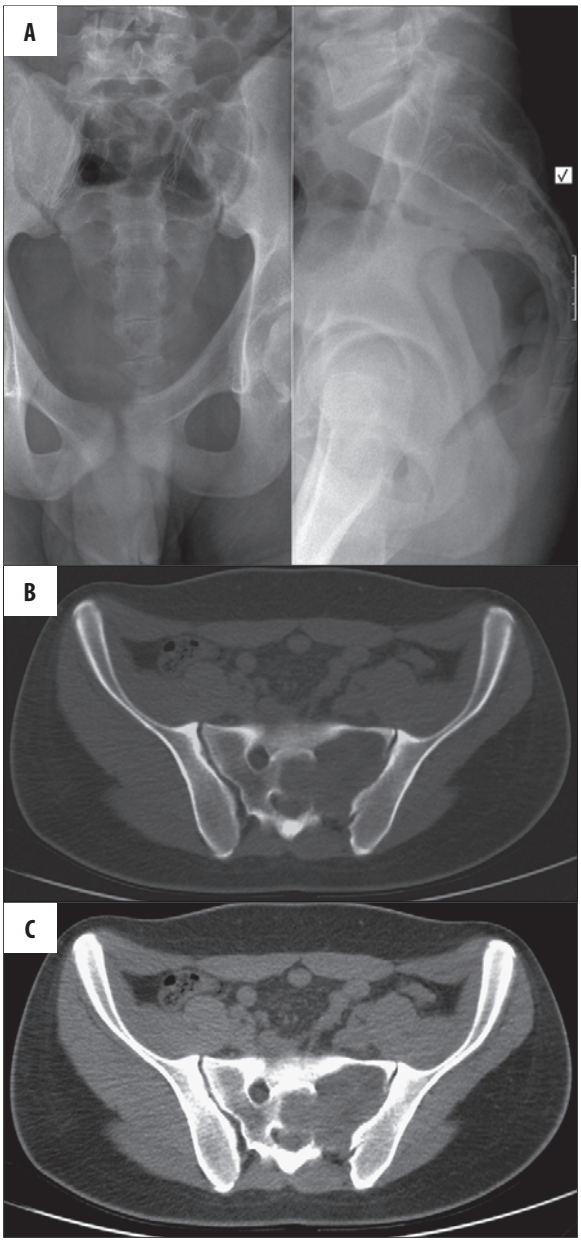


Figure 4. Patient Z.A. (**A**) Plain film. Osteolytic focus in the left lateral mass of the sacrum, difficult to assess due to gas in the bowels. (**B,C**) CT. ABC: mass in the sacrum on the left, without the signs of malignancy, with only a few fluid levels (**B** – bone window, **C** – soft tissue window).

areas depending on the content of the cysts [2,3,6,15]. After intravenous contrast medium administration, ring-like enhancement is observed within the septa, as well as within the solid elements of the cysts [2]. Scintigraphy, not included in our study, is a sensitive but non-specific method, in which intensive radioligand uptake is observed on the periphery of the lesion, with low uptake in the central part [1,7,16].

Conventional radiography is considered sufficient for the diagnosis of typical ABC, whereas CT and MRI can extend the diagnostics in unclear cases (Figure 3A–C), with location atypical or difficult to access by plain x-ray (Figure 4A–C) [1,2,7,16]. In our material, the accuracy of radiography-based diagnoses was only 55.5%, it was higher For CT – 63%, and the highest for MRI – 75%. However, MRI was performed only in 8 cases. The diagnosis was accurate in 50% of cases examined with US. The accuracy is quite high in view of some opinions questioning the value of US scans in ABC diagnostics [7]. The accuracy markedly increases if all four imaging modalities are used, reaching 72.2%.

What was the reason for low diagnostic efficacy, especially in case of radiography, in our material? In most cases (65.3%), the radiograms revealed a single osteolytic focus

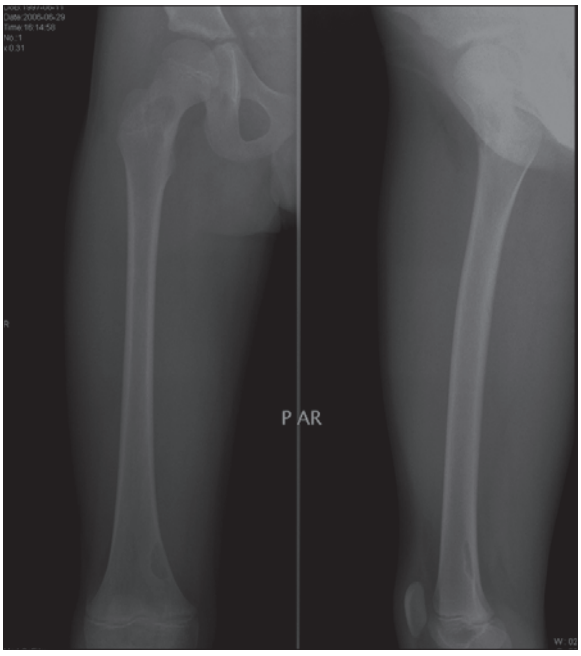


Figure 5. Plain film. Unclear osteolytic focus in the proximal metaphysis of the femur, suspicious of a malignant lesion on MRI (cortical fibrous defect in the distal metaphysis).

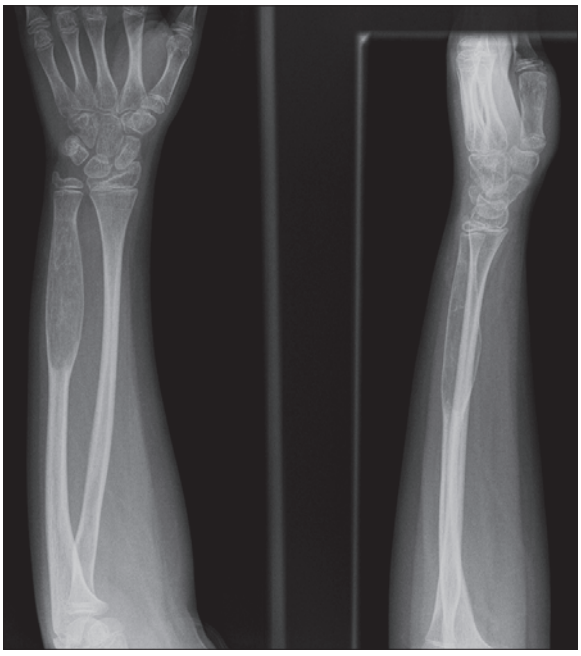


Figure 6. Plain film. Suspicion of a simple cyst: homogeneous osteolytic lesion in the diaphysis of the ulna, bone expansion.

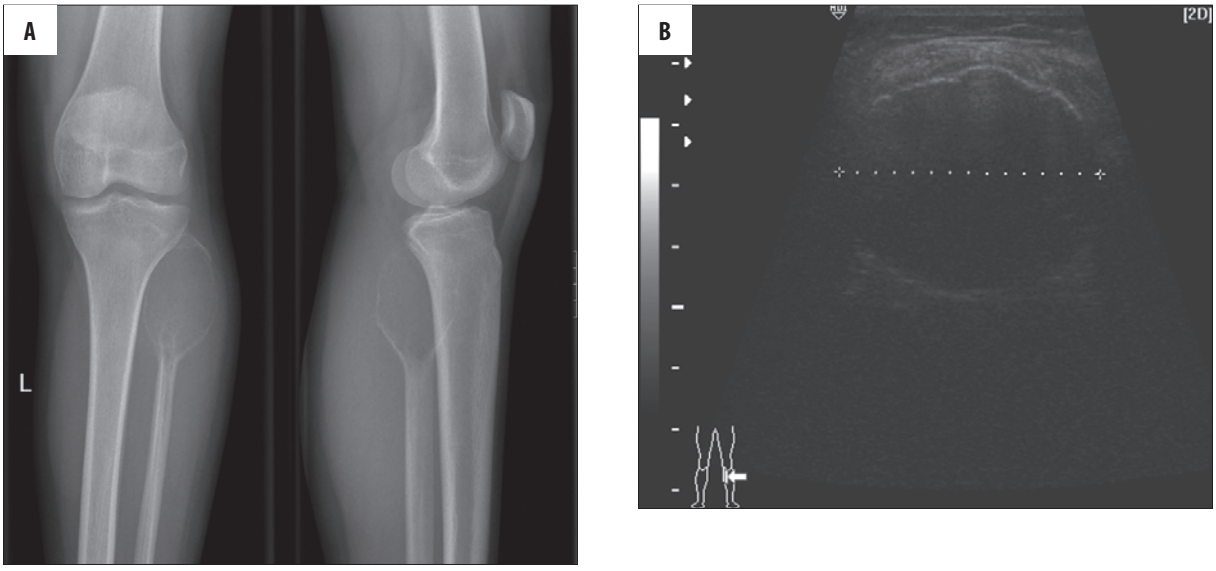
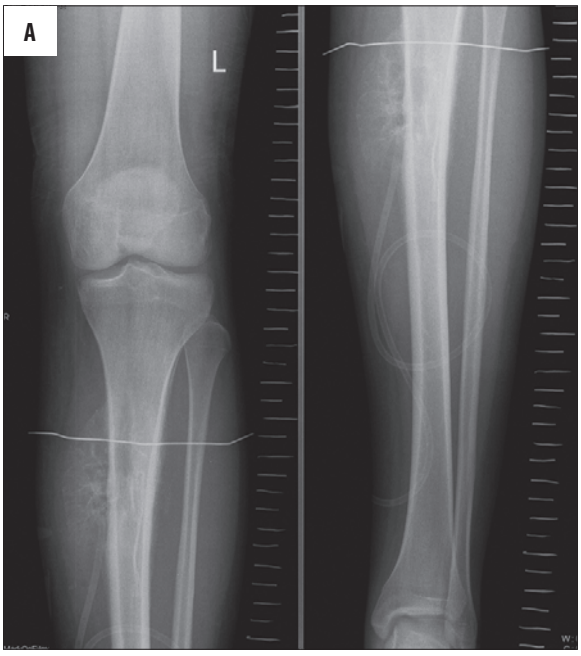


Figure 7. Patient M.J. (**A**) Plain film. ABC in the proximal metaphysis of the fibula: osteolytic focus, bone expansion, the cortex is thinned and broken. (**B**) US. Cystic lesion.

rather than policyclic, cyst-like lesions (Figures 5,6). In over 50% of cases (55.5%) no septa were seen within the osteolytic foci, and in 21% signs of malignancy, such as obscured outline of the osteolytic lesion, disrupted cortical layer accompanied by ill-delineated periosseous mass and periosteal reactions malignant in character, i.e. periosteum disruption and Codman's triangles were observed.

In 1985, R. Capana and D. A. Campanacci described 5 morphologic types of ABC on the basis of radiograms [16]:

- type I – the cyst occupies the central part of the bone, usually the metaphysis whose outline remains unchanged or is slightly expanded;
- type II – the cyst extends to the whole width of the bone, with considerable expansion, thinned cortical layer, often at the borderline between the metaphysis and diaphysis of thin long bones such as the fibula, radius and ulna;
- type III – the cyst is located eccentrically in the metaphysis, sometimes extending slightly to the cortical layer;
- type IV – the cyst develops subperiosteally and may cause superficial erosions of the cortical layer; this type is observed rarely in long bone diaphyses;
- type V – localization at the borderline between the metaphysis and diaphysis of long bones, subperiosteal and intraosseous; the lesion causes detachment and displacement of the periosteum towards the soft tissue and destroys the cortical layer.



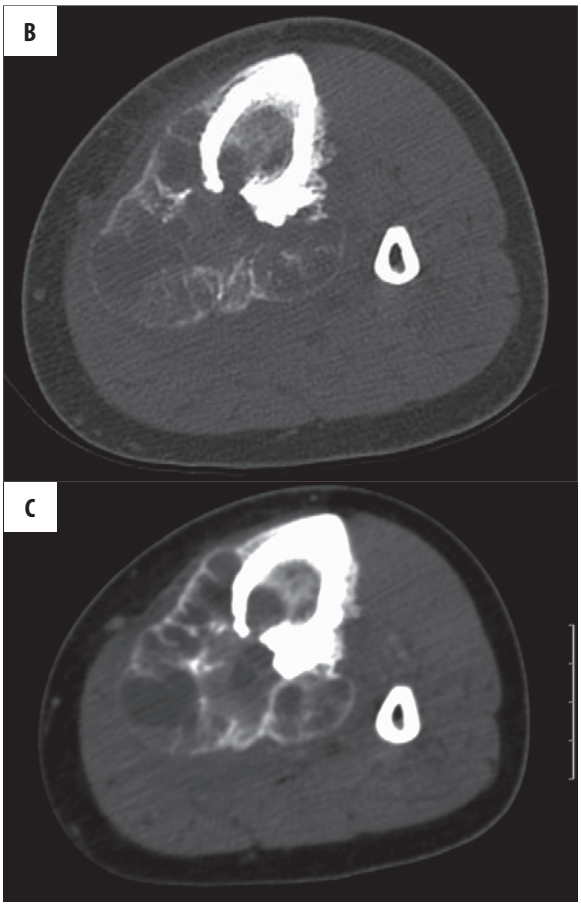


Figure 8. Patient O.P. (**A**) Plain film. Suspicion of a malignant lesion: irregular osteolytic defect, ill-defined periosseous mass, Codman's triangle, spicular periosteal reaction. (**B,C**) CT. ABC: sclerotic rim, septa, fluid levels (**B** – bone window, **C** – soft tissue window).

M. K. N. Anand distinguishes four phases of ABC development, defined on the basis of the cyst relation to the adjacent structures [7]:

- phase I, initial, in which the osteolytic lesion localized inside the bone is well-delineated by the reactive bone layer without contact with the periosteum;
- phase II, of active growth, involves rapid bone destruction and subperiosteal expansion of the lesion; periosteal reaction is still incomplete;
- phase III, mature, is characterized by the presence of a sclerotic layer on the periphery (reactive periosteal bone formation) and osseous septa within the cysts, yielding typical presentation of ABC;
- phase IV is the recovery phase with progressive calcification and ossification of the cyst.

It seems justifiable to state that radiographic presentation of ABC is dependent both on the cyst type and on the phase in which it is examined. While the type has lower diagnostic significance, the stage of cyst development can considerably affect the diagnosis because only phases III and IV allow to make diagnostic suggestions with most certainty. In our material, phase I and II lesions seemed to predominate (Figure 7A,B), as indicated in most cases by single osteolytic foci (65.3% on x-ray, 61% on CT), bone expansion (86% on x-ray, 61% on CT), cortical layer destruction (26% on x-ray, 56,3% on CT), lack of septa on x-ray in 55.5% of cases, lack of fluid levels on CT in 73%, on US in 65% and in 50% of cases on MRI.

Similarly, radiological signs of malignancy observed in our material can be explained by the dependence of presentation of the phase of ABC development [2,16]. In phase II, characterized by active growth, the lesion penetrates both the cortical layer of the bone and the subperiosteal region. Imaging studies show disruption of the cortical layer with obscured intraosseous borderline, a soft tissue tumor, i.e. subperiosteal periosseous mass of the cyst, ill-delineated on the periphery, because full periosteal reaction has not developed yet and the periosteum is not calcified enough to be visualized in the radiogram [16,18]. It also seems that inhomogeneous calcification of the periosteum can be interpreted as its disruption – no evidence of actual disruption of the periosteum by the cystic mass has been found in the available literature. Because of its properties, CT visualizes the periosteum better than radiography [18] (Figure 8A–C). Atypical contrast enhancement in CT and MRI, as well as increased flow in US scans with Power Doppler option, interpreted in our study as a sign of malignancy, was probably associated with the presence of more solid elements in the examined cysts.

The results concerning the general characteristics of ABC are mostly consistent with literature data. In our material, the average age of patients at commencement of the diagnostics was 12 years, the highest incidence was observed in the 10–19 age group (67%). According to most authors, ABC in 50–70% become manifest in the second decade of life and the mean age at which they are diagnosed is 13–17 years [1,4]. Most publications report a slight predominance of female patients [1,4,7], whereas boys predominated in our material.

Most frequent localization of the lesions in long bones of the lower extremities (43%) with metaphyseal location

(56.6%) and in flat bones (25%) is confirmed by the literature [1,4,7]. In contrast to other publications [1,7], reporting relatively frequent localization of ABC within the posterior parts of the vertebral bodies, in our material there was a single case of such localization.

An ABC may develop in a healthy bone, or in ca. ⅓ of cases as a part of, or a lesion secondary to, previously existing bone tumors [7]. In our material, primary cysts were diagnosed in 65.3% of cases.

Solid form of ABC is a rare variant [1–3,7], no such cases were found in our material.

According to some authors, the diagnosis of ABC can be established on the basis of pathognomonic x-ray and CT images, and in dubious cases open biopsy is necessary to exclude frequently occurring concomitant tumors. Other authors believe that histological diagnosis is recommended in each case to make sure there is no malignant process [1,5].

The overall diagnostic accuracy amounted to 72.2%. A typical ABC usually poses no diagnostic problems and some authors estimate the efficacy of imaging modalities at nearly 100% [2,7]. On the other hand, some authors regard ABC diagnostics as difficult with malignant tumors and simple cysts as the most common incorrect diagnoses [4,5,16].

Conclusions

1. The diagnosis of aneurysmal bone cysts based on plain radiography is possible in cases with typical radiological signs.

2. Application of other imaging methods (CT, US, MRI) significantly increases the percentage of correct diagnoses.

3. Lack of fluid levels and osseous septa does not exclude the diagnosis of ABC.

4. Imaging features of malignancy do not exclude the diagnosis of ABC.

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