Plain Radiologic Assessment of Unicameral and Aneurysmal Bone Cyst: Differential Diagnosis

Estudio radiológico simple en quiste óseo simple y aneurismático: Diagnóstico diferencial

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Abstract

Objective To describe our experience in the differential diagnosis of unicameral and aneurysmal bone cysts using plain radiography.

Patients and method We included patients under 20 years of age with radiographs of histologically-confirmed unicameral or aneurysmal bone cysts found on the Chilean National Bone-Tumor Registry. The radiographs were evaluated by two experienced radiologists. We compared the demographic variables of the patients, and the clinical and radiological variables of both tumors..

Results A total of 97 patients met the inclusion criteria, 65% of whom had simple bone cysts, and 35%, aneurysmal bone cysts. No differences were found regarding age, the bone affected, the size of the lesion, bone expansion, nor cortical thinning. Gender, clinical presentation, cortical interruption, and location of the lesion on the longitudinal and transverse bone axes are parameters that could be used in the differential diagnosis of both cysts.

Discussion According to the literature, unicameral and aneurysmal bone cysts are frequent benign lesions that are difficult to differentiate merely through plain radiographs. Magnetic resonance imaging enables a better anatomical characterization and provides sensitivity and specificity to the diagnosis. However, its availability is limited, and it should be preceded by plain radiography.

Keywords
- differential diagnosis
- bone cyst
- aneurysmal bone cyst
- radiography
- bone neoplasms

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Conclusion Plain radiography is still the initial imaging study of choice in patients with clinical suspicion of bone tumor, even in those centers where magnetic resonance imaging is available. Certain demographic and radiological characteristics guide physicians in the differential diagnosis of unicameral and aneurysmal bone cysts.

Resumen

Objetivo Dar a conocer nuestra experiencia en el diagnóstico diferencial de los quistes óseos simple y aneurismático mediante estudio radiológico simple.

Materiales y métodos Se incluyeron pacientes menores de 20 años con radiografía de quiste óseo simple o aneurismático histológicamente confirmado pertenecientes al Registro Nacional de Tumores Óseos. Las radiografías fueron analizadas por dos radiólogos experimentados. Se compararon las variables demográficas de los pacientes, y las variables clínicas y radiológicas de ambos quistes.

Resultados Un total de 97 pacientes cumplieron los criterios de inclusión (65% presentaba quistes óseos simples y 35%, aneurismáticos). No se observaron diferencias en cuanto a la edad, al hueso comprometido, al tamaño de la lesión, a la expansión ósea, ni al adelgazamiento cortical. El género, la forma de presentación, la presencia de interrupción de la cortical, y la localización de la lesión en los ejes longitudinal y transversal del hueso son parámetros que podrían ser útiles en el diagnóstico diferencial de ambos quistes.

Discusión Los quistes óseos simple y aneurismático son lesiones benignas frecuentes que, de acuerdo con la literatura, serían difíciles de diferenciar únicamente con radiografía simple. La resonancia magnética permite una mejor caracterización anatómica, y aporta sensibilidad y especificidad al diagnóstico. Sin embargo, debe ser precedida por la radiografía simple, y su disponibilidad es limitada.

Conclusión Aun en centros con disponibilidad de resonancia magnética, la radiografía simple sigue siendo el estudio inicial de elección en el diagnóstico de tumores óseos. Ciertas características demográficas y radiográficas permiten orientar el diagnóstico diferencial inicial entre el quiste óseo simple y el aneurismático.

Palabras clave
- diagnóstico diferencial
- quiste óseo
- quiste óseo aneurismático
- radiografía
- tumores óseos

Introduction

Bone injuries are very common, especially in children. The literature[1,2] estimates that up to 42% of all bone lesions occur during the first two decades of life. The most frequent benign lesions in children are: osteochondroma, non-ossifying fibroma, Langerhans cell histiocytosis, unicameral bone cyst (UBC), and aneurysmal bone cyst (ABC).[2]

Imaging examinations are essential for the initial diagnosis of bone tumors, for they guide clinical decisions regarding the management of this type of patient. Plain radiography is the most useful imaging modality to establish the initial differential diagnosis of bone tumors, and it is also widely available.[3,4]

Unicameral bone cysts are benign lesions formed by fluid-filled cavities that tend to expand and weaken the bone locally. They represent 3% of biopsied bone tumors, ad are slightly more frequent in men (in a proportion of 2.5:1).[5] In most cases, they originate in the metaphyses of the long bones adjacent to the physis and, as the individual grows, they tend to move away from the physis, migrating towards the metaphyseodiaphyseal region or even towards the diaphysis. Unicameral bone cysts rarely cross the physis into the epiphysis, so epiphyseal involvement is unusual.[6,7] They affect growing patients, up to 85% of the patients are under 20 years of age,[5,8] and the age group between 10 and 20 years is the most affected.[7] The bones most frequently compromised are the humerus and the proximal femur.[5,8] Other locations described are the calcaneus, the ilium, the distal radius, and the patella.[7]

Unicameral bone cysts usually present as fractures in pathologic bones.[5] In certain cases, a fragment of the wall of the fractured cyst is deposited in its cavity, which is known as the "fallen fragment sign" or "fallen leaf sign".[5,9] If there has been no fracture, they can present as an osteolytic lesion well delimited by a sclerotic margin, which can grow and expand the juxta-epiphyseal metaphysis of the bone.[5] Regarding the radiological presentation of UBCs, they are typically described as a central lesion, which does not cross the cortical bone or form extraosseous lesions.[10] Magnetic resonance imaging enables the precise delimitation of the collection, and, in the event of a fracture, to determine a level.[5] The differential diagnosis mainly includes UBC and fibrous dysplasia.[5]

Aneurysmal bone cysts are benign bone lesions that may present clinically with pain, increase in volume, or as an expansive mass, and the presentation of this lesion with pathologic bone fracture is unusual.[7,11,12] They have a low incidence among the population (0.14 to 0.32 per 100 thousand individuals), and are more frequent in the first two decades of life.[11–15] They represent approximately 1% of all bone tumors.[16] The most frequent sites are the humerus, the femur, the tibia, and the fibula. Other sites that may be affected are the skull and the posterior elements of the spine.[11,12] Regarding their location in the bone, they are generally observed in the metaphysis and, like the UBCs during growth, the physis tends to move away from the lesion, acquiring a metaphyseodiaphyseal location. It has been reported that up to 10% are diaphyseal.[17] Aneurysmal bone cysts are typically described as eccentric lesions, but they can also be concentric or subperiosteal,[11] resulting in bone expansion. The management of these bone tumors is usually surgical, with the main objective of completely eradicating the lesion in order to minimize the risk of recurrence.[11,12]

The literature indicates that ABCs can be difficult to differentiate from UBCs based exclusively on the information provided by radiography.[12] Magnetic resonance imaging compared with conventional radiography has shown to have greater specificity in the diagnosis, and the combination of both examinations would improve the sensitivity, specificity and positive predictive value.[18] However, both studies are not always available in clinical practice. In this context, it may be especially relevant to know the distinctive radiographic characteristics of both lesions, in order to facilitate the diagnosis and contribute to the appropriate subsequent clinical conduct. The objective of the present publication is to show our experience in the differential diagnosis of UBCs and ABCs in plain radiological studies.

Materials and Methods

The Chilean National Bone Tumor Registry (Registro Nacional de Tumores Óseos, RENATO, in Spanish) was retrospectively analyzed in search of radiographs of patients under 20 years of age with a histologically-confirmed diagnosis of UBC or ABC. The RENATO was initiated and coordinated by Drs. Juan Fortune H., Fernán Díaz B., and Martín Etchart K., who were then joined by Dr. Jaime Paulós A., between 1959 and 1999 at Pontificia Universidad Católica, Santiago, Chile. The radiological analysis was performed by two experienced radiologists (AB, CG). To compare the findings of UBC and ABC, the following parameters were tabulated: gender, age group ($<$ 6 years, 6 to 10 years, and 11 to 20 years), affected bone, tumor size ($<$ 2 cm, 2 to 5 cm, 6 to 10 cm, and $>$ 10 cm), location of the tumor in the longitudinal and transverse axes of the bone, presence of bone expansion, thinning and interruption of the cortex, periosteal reaction, and soft-tissue involvement. Numbers and percentages were used to express the categorical variables.

Results

A total of 98 patients met the inclusion criteria, but 97 were included in the analysis. One patient in whom an apparent error was detected in the transcription of the pathological diagnosis was excluded from the analysis, since the correct diagnosis could not be confirmed because the original sample was not available. It should be noted that no information regarding age was found in the records of 6 patients (4 with UBC and 2 with ABC), so only the age of the remaining 92 cases was analyzed. In four cases (teo with UBC and two with ABC), it was not possible to determine whether they were eccentric or central lesions, since there was only one projection, so they were not included in the analysis of this characteristic. Cases with a fracture and an increase in the volume of the soft tissues in the area of the injury were considered pathologic bone fractures.

In total, 63 (65%) patients with UBC and 34 (35%) with ABC were identified. In the analysis by age group, both UBCs and ABCs were more frequent in the segment between 11 and 20-years. Unicameral bone cysts were more common among men (65%), while ABCs were more common among women (56%). According to the location in the bone, UBCs affected the large bones in 58 patients (92%) and ABCs affected the long bones in 26 patients (76%), and the most affected bones were the humerus and the femur. Unicameral bone cysts were the only ones to compromise the calcaneus and ABCs were the only ones to compromise the vertebrae and scapula.

Metaphyseodiaphyseal involvement was observed in approximately half of the cases of both tumors (►**Figs.1–5**, **6–8**). Only one patient (3%) with an ABC presented simultaneous involvement of the diaphysis, metaphysis, and epiphysis. Exclusive involvement of the diaphysis was more frequent in UBCs than in ABCs (►**Figs. 2**,**9**).

The size was similar in both lesions. In total, 51% (32 patients) of the UBCs measured between 2 cm and 5 cm, and 35% (22 patients), between 6 cm and 10 cm. On the other

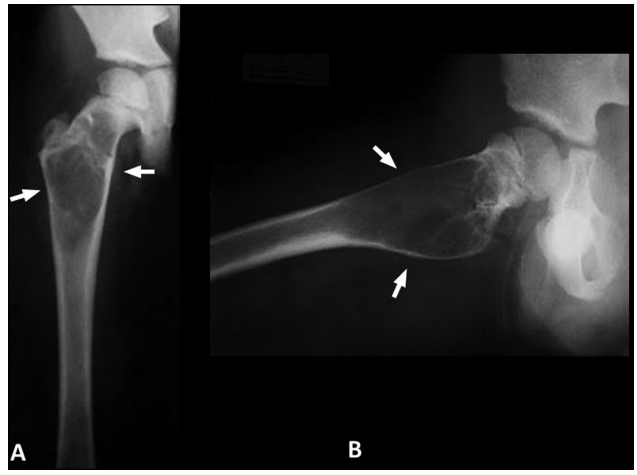

Fig. 1 12-year-old boy. Radiograph of the right hip in anteroposterior (AP) (A) and Lowenstein (**B**) views, with a unicameral bone cyst in the right femur that involves the proximal metaphyseodiaphyseal region, with signs of bone expansion and endosteal cortical thinning, without cortical perforation (arrows).

Fig. 2 7-year-old boy. Radiograph of the right humerus, in AP view, with a unicameral bone cyst that compromises the proximal diaphysis, associated with deformity and bone curvature, and without clinical or radiological signs of fracture (arrows).

Fig. 3 16-year-old boy. Right-leg X-ray, in AP (**A**) and lateral (**B**) views, with a unicameral bone cyst in the right fibula that involves the proximal metaphyseodiaphyseal region, showing bone expansion and well-defined sclerotic margins (arrows).

hand, regarding the ABCs, 56% (19 patients) measured between 2 cm and 5 cm, and 35% (12 patients), between and 6 cm and 10 cm.

Analyzing the location in the bone, we observed that UBCs were present in almost every case (97% – 61 patients) as a central lesion (►**Figs.1–3**, **5**,**6**), while 23% (8 patients) of the ABCs were eccentric (►**Fig.8**).

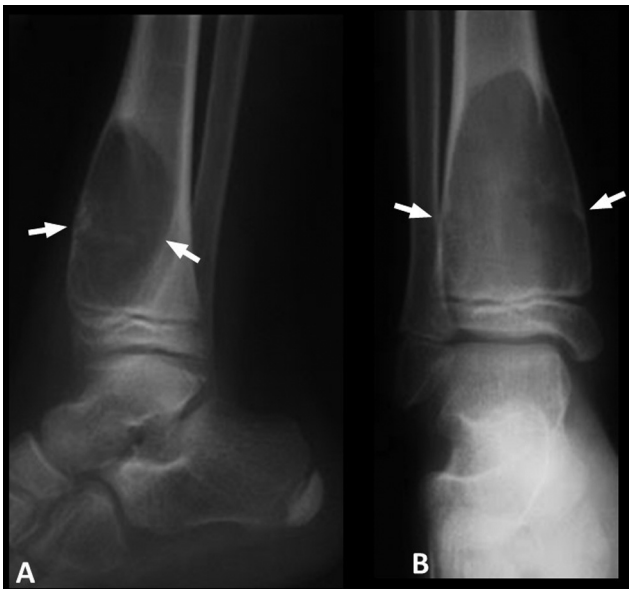

Fig. 4 9-year-old girl. Radiograph of the right ankle in AP (**A**) and lateral (**B**) views, with an expansive unicameral bone cyst in the right tibia, which thins the cortex and compromises the distal metaphyseodiaphyseal region, without signs of fracture. The lesion is central in the mediolateral axis of the bone; however, it is eccentric in the AP axis (arrows).

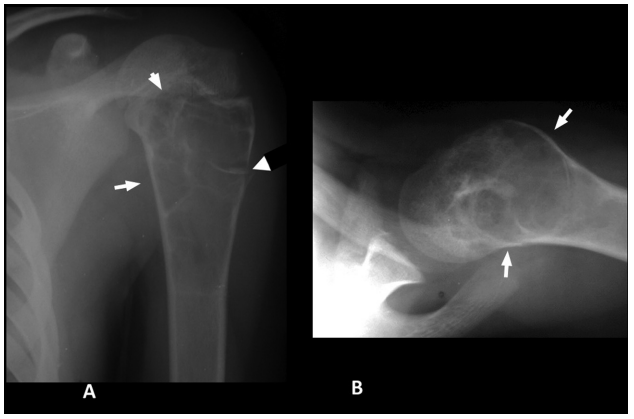

Fig. 5 13-year-old girl. Radiograph of the right humerus in AP (**A**) and lateral (**B**) views, with an aneurysmal bone cyst in the diaphysis of the right humerus (arrows) associated with a fracture and with the sign of the fallen fragment (arrowhead).

The presence of fractures was more frequent in the cases of UBC than in those of ABC (►**Figs. 2**, **5**,**10**).

Bone expansion and cortical thinning were observed in most lesions (►**Figs.3**, **5**, **11**, **8–10**). Cortical disruption was more frequent in ABCs than in UBCs (►**Fig. 8**). Periosteal reaction was an infrequent finding in both lesions, only 3% (2 patients with UBC and 1 patient with ABC) of the cases UBC and ABC presented it. There was no soft-tissue involvement in the cases studied, except in one (1%) case of ABC, with posttraumatic volume increase (►**Fig. 10**).

The details of the results obtained are described in ►**Tables 1** and **2**.

Fig. 6 18-year-old male. Radiograph of the right foot, in oblique view, with an aneurysmal bone cyst in the third metatarsal, compromising the distal metaphyseodiaphyseal region (arrows), as an expansive lesion, with cortical thinning.

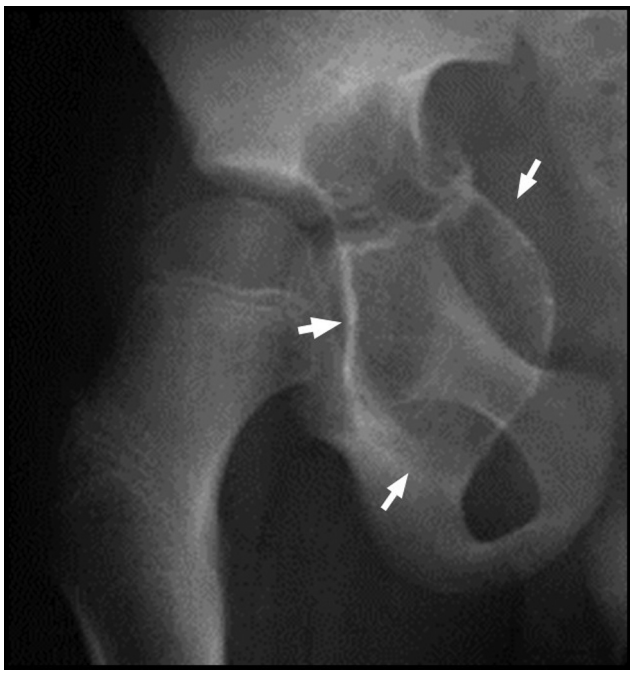

Fig. 7 9-year-old boy. Pelvic X-ray, in AP view, with an aneurysmal bone cyst that mainly involves the acetabulum and the upper branch of the pubis, expansive in nature, with thinning of the cortex (arrows).

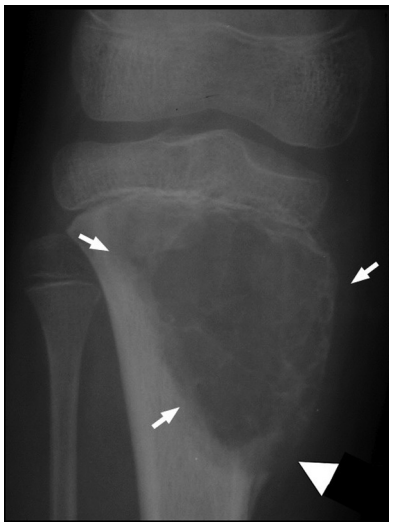

Fig. 8 14-year-old boy. Right-knee X-ray, in AP view, with an aneurysmal bone cyst involving the proximal metaphyseodiaphyseal region of the tibia, eccentric in location, with bone expansion, cortical thinning, and cortical interruption (arrow).

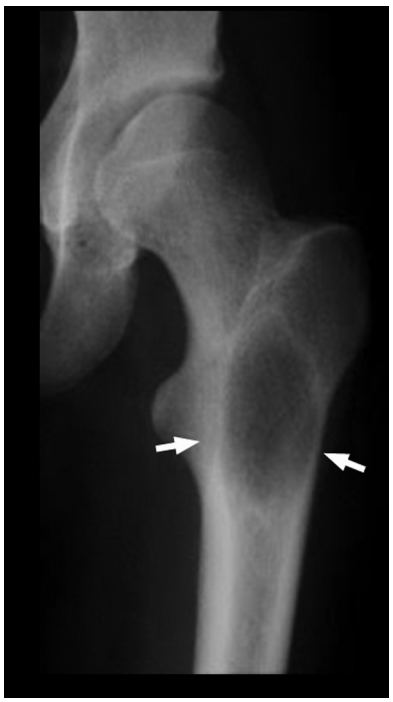

Fig. 9 13-year-old boy. Left-hip radiograph, in AP view, with an aneurysmal bone cyst in the intertrochanteric and subtrochanteric regions of the proximal femur, as a phenomenon of "migration" of the lesion towards the diaphysis (arrows).

Discussion

Unicameral bone cysts were more frequent among males between 11 and 20 years of age, and ABCs, among women of the same age group, which coincides with the reports in the literature.[5,7,8,11–15] Regarding the compromised bone, in both types of cyst, the most frequently-affected bone was the humerus, followed by the femur, which is also similar to the reports in other reviews.[5,8,11,12] Up to 15% of involvement of the spine has also been described in ABCs,[7] which also was reflected in our experience, since 9% (3 patients) of the lesions were located in the spine, with involvement of the vertebral body, the pedicles, and the spinous processes.[11,12] Likewise, involvement of the calcaneus and iliac bones has been described[19] in cases of UBC, which was reflected in our results.

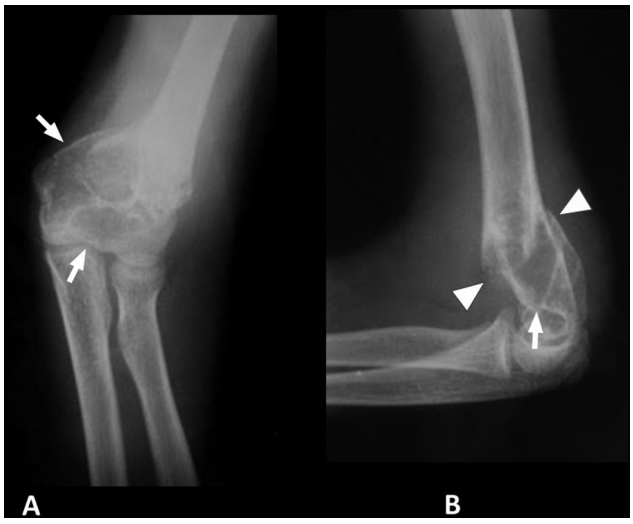

Fig. 10 13-year-old girl. Left-elbow X-ray, in AP (**A**) and lateral (**B**) views, with an aneurysmal bone cyst in the left humerus, involving the distal metaphyseal region (arrows), associated with a supracondylar fracture, with posterior angulation, displacement, and increased volume of the soft parts (arrow heads).

Fig. 11 6-year-old girl. Radiograph of the right scapula, in AP views, with an aneurysmal bone cyst in the right scapula, manifested as an expansive lesion, with cortical thinning, without fracture (arrows).

Table 1 General characteristics of 97 cases of UBC and ABC

		UBC (N = 63)		ABC (N = 34)	
		N	%	N	%
Age	< 6 years	11	19%	2	6%
	6 to 10 years	15	25%	12	38%
	11 to 20 years	33	56%	18	56%
Gender	Male	41	65%	15	44%
	Female	22	35%	19	56%
Compromised bone	Humerus	31	49%	11	32%
	Femur	16	25%	5	15%
	Tibia	5	8%	5	15%
	Fibula	5	8%	3	9%
	Calcaneus	3	5%	–	–
	Metacarpal	1	2%	1	3%
	Ulna	1	2%	2	6%
	Pelvis	1	2%	2	6%
	Scapula	–	–	1	3%
	Metatarsal	–	–	1	3%
	Vertebra	–	–	3	9%
	Epiphysis	–	–	–	–
Location in the longitudinal axis	Metaphysis	1	2%	–	–
	Diaphysis	28	44%	8	24%
	Epiphysis + metaphysis	–	–	1	3%
	Diaphysis + metaphysis	30	48%	17	50%

Table 1 (*Continued*)

		UBC (N = 63)		ABC (N = 34)	
		N	%	N	%
	Epiphysis + metaphysis + diaphysis	–	–	1	3%
	Femoral neck	–	–	1	3%
	Calcaneus	1	2%	–	–
	Inferior angle of the scapula	–	–	1	3%
	Vertebral body and pedicles	–	–	2	6%
	Ischium	–	–	2	6%
	Pedicle and the spinous process	–	–	1	3%
	Iliac bone	1	2%	–	–
Location in the transversal axis	Central	59	97%	23	77%
	Eccentric	2	3%	7	23%

Abbreviations: ABC, aneurysmal bone cyst; UBC, unicameral bone cyst.

Table 2 Radiological characteristics of 97 cases of UBC and ABC

		UBC (N = 63)		ABC (N = 34)	
		N	%	N	%
Fracture	Yes	34	54%	13	38%
	No	29	46%	21	62%
Size	< 2 cm	3	5%	1	3%
	2 to 5 cm	32	51%	19	56%
	6 to 10 cm	22	35%	12	35%
	> 10 cm	6	10%	2	6%
Periosteal reaction	Yes	2	3%	1	3%
	No	61	97%	33	97%
Fallen fragment	Yes	–	–	–	–
	No	63	100%	34	100%

Abbreviations: ABC, aneurysmal bone cyst; UBC, unicameral bone cyst.

Regarding the location in the bone in its longitudinal and transverse axes, both lesions have been described[6,7,17] more frequently in the metaphysis with an apparent migration towards the metaphyseodiaphyseal region during growth. In our experience, involvement of the metaphyseodiaphyseal region was observed in 48% (30 patients) of UBCs and in 50% (17 patients) of ABCs. There was also a high percentage of diaphyseal involvement, which was higher among the cases of UBC. Although it has been described[17] that ABCs can have a diaphyseal location in up to 10% of the cases, our results show a higher percentage, which is probably explained by the migration of these lesions towards the diaphysis during growth.

Regarding their location in the transverse axis of the bone, UBCs are typically described as central, and ABCs, as eccentric, although the latter may also have central or subperiosteal location.[6,11] In our experience, although most of the cases of UBC and ABC were central, there was greater eccentric engagement in ABCs.

Regarding their radiological appearance, both lesions produce bone expansion and cortical thinning, which

Table 3 Relevant features relevant for the diagnoses of UBC and ABC

	UBC	ABC
Gender	More common among men	More common among women
Presentation	Pathologic bone fracture	Pain or swelling, unusual fracture
Location in the longitudinal axis	Metaphyseodiaphyseal and diaphyseal regions	Metaphyseodiaphyseal and diaphyseal regions
Location in the transversal Axis	Central	Eccentric
Cortical disruption	Usually does not present cortical disruption	Cortical disruption is more common

Abbreviations: ABC, aneurysmal bone cyst; UBC, unicameral bone cyst.

coincides with our results, as well as the higher percentage of cortical interruption in ABCs. The low percentage of periosteal reaction found in ABCs is striking, since previous reviews[2,12] indicate that this is a more frequent finding. Similarly, this finding is not described regarding UBCs, unless there is a fracture.[10] However, we have reported the aforementioned characteristic in 2 cases (3%) in which there was no fracture. The sign of the fallen fragment in UBCs was not demonstrated with certainty, probably because most of the examinations were probably obtained in the decubitus position, as is usually the case in pediatric patients.

Therefore, both lesions share similar characteristics in terms of age, compromised bone, size, bone expansion, and cortical thinning. Some parameters that would enable the establishment of the differential diagnosis between UBCs and ABCs by means of plain radiological studies are gender, form of presentation, location in the longitudinal and transverse axes of the bone, and the presence of interruption of the cortical bone. Unicameral bone cyst is frequent among men, its onset is usually due to a pathologic bone fracture, it tends to be located in the metaphyseodiaphyseal region or the region of the diaphysis, it is central, and it generally does not interrupt the cortical bone, while ABC is more frequent among women, it presents with an increase in volume and pain, with fracture being an unusual presentation, it is located mainly in the metaphyseodiaphyseal region with less isolated compromise of the diaphysis, the interruption of the cortical is more frequent and, in a higher percentage of cases, it is eccentric. These characteristics may be useful for the radiologist when it comes to differentiating both lesions based exclusively on the findings of plain radiography (►**Table 3**).

Conflict of Interests
The authors have no conflict of interests to declare.

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