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MR imaging of simple bone cysts in children: not so simple

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Abstract *Objective*. The unicameral bone cyst (UBC) is a common cystic bone lesion seen in children. We review and summarize its MR findings, focusing on their appearance following contrast enhancement, and correlating them to known histologic features of UBC.
Subjects and methods. A retrospective review of 20 cases (13 boys, 7 girls; age range, 1–17 years; mean age, 8.9) diagnosed as UBC was conducted. Clinical histories, radiographic and MR features, follow-up data, and available pathologic findings were noted.
Results. At initial presentation 18 (90 %) of the patients with UBC had a history of acute or remote pathologic fracture. Cysts with history of fracture displayed heterogeneous fluid signals on T1- (n = 9, 50 %), and T2-weighted (n = 15, 83 %) MR images. Gadolinium-enhanced images were obtained in 15 of the 18 UBC cases with history of previous fracture. All showed enhancement with focal, thick peripheral, heterogeneous, or subcortical patterns. Focal nodules of homogeneous enhancement (diameter > 1 cm) within the UBC (n = 5) correlated with areas of ground-glass opacification on plain film. Other interesting MR features were fluid-fluid levels (n = 11), "fallen-leaf" sign (n = 1), soft-tissue changes (n = 2), and detection of septations not seen on plain film (n = 2).
Conclusion. UBCs frequently appear complicated on MR imaging, with heterogeneous fluid signals and regions of nodular and thick peripheral enhancement related to previous pathologic fracture and early healing.

Introduction

Simple or unicameral bone cysts (UBCs) are common benign lesions in children, of unknown cause, typically arising in the proximal metaphyses of tubular bones adjacent to the growth plate and are generally discovered incidentally or following pathologic fracture [1].

Magnetic resonance (MR) imaging has not been extensively used in the diagnosis and evaluation of UBCs because of their characteristic radiographic appearance [1]. On plain film, these cysts are usually central, elongated radiolucent lesions with cortical thinning, minor osseous expansion, and well-defined margins [1]. They may sometimes be difficult to diagnose on plain film, since they can appear multilocular, occur in uncommon sites, or show other radiographic features arising from early healing or previous pathologic fracture [1, 2]. In these cases MR imaging may be obtained as diagnostic considerations can include enchondroma, aneurysmal bone cyst, fibrous dysplasia, nonossifying fibroma, or rarely telangiectatic osteosarcoma.

Previous MR descriptions of UBC have consisted primarily of single case reports, emphasizing the prolonged T1 and T2 relaxation times and simple fluid con-

tent of the lesions [1, 2–5], although isolated cases with internal hemorrhage and fluid-fluid levels have been reported [1, 6]. To our knowledge, the typical MR features of the UBC at presentation, gadolinium-enhancement characteristics, and appearance following partial healing or pathologic fracture have not been described in a larger series.

Subjects and methods

We conducted a retrospective review of 20 cases (13 boys, 7 girls; age range 1–17 years; mean age 8.9 years) diagnosed as UBC from 1990 to 1998. MR images were available in every case.

History before imaging

At the time of referral, UBCs were divided into two groups based on clinical history. Lesions with a history of acute or remote pathologic fracture were classified as complicated, those without a history of pathologic fracture as uncomplicated. Two of the complicated lesions had undergone treatment before referral. The mean interval between previous treatment and time of imaging was 4.5 months.

Imaging

At the time of referral, all patients underwent both X-ray and MR evaluation. The interval between imaging modalities was no more than 1 month. Plain-film images were examined for previously described prognostic features [7, 8], including the presence of normal bone between the lesion and the physis, ground-glass opacification, septations, and periosteal reaction.

Multiplanar spin-echo T1 and T2 ($n = 20$, 100 %) and T1 fat-suppressed gadolinium-enhanced ($n = 16$, 80 %) MR sequences were obtained using GE Signa (General Electric, Milwaukee, Wis.) or Siemens SP 4000 (Siemens, Erlangen, Germany) machines operating at 1.5 T. T1- and T2-weighted images were examined for signal intensity and pattern of homogeneity. Signal intensities less or greater than those of adjacent skeletal muscle were considered low or high, respectively. The enhancement patterns following gadolinium infusion (0.1 mmol/kg) were classified as thick- or thin-rim, septal, focal (large nodules, diameter > 1 cm; small loci, diameter < 1 cm), heterogeneous, or subcortical. The presence of one or more fluid-fluid levels, internal hemorrhages, septations, and adjacent soft-tissue changes were also noted.

Treatment and follow-up

The diagnosis of UBC was established through intraoperative aspiration of clear or blood-tinged fluid and cystogram. In questionable cases, such as poor filling on cystogram, open biopsy was performed ($n = 2$). Routine cytology was only conducted in two cases, in which the lesions were poorly demarcated or recurrent or were suspected of containing an aneurysmal component. Treatment was provided to 17 patients and consisted of steroid injection ($n = 3$), autologous bone-marrow injection ($n = 12$), and curettage with autologous bone marrow injection ($n = 2$). In the remaining three cases no treatment was given, as intraoperative examination showed healing.

Follow-up was conducted by plain radiography. The earliest time at which a radiological response could be reliably determined was 3 months; however; mean follow-up was 19 months. After follow-up, cysts were classified as showing complete healing (consolidation and cortical thickening), incomplete healing (consolidation with persistent areas of osteolysis), recurrence (large areas of osteolysis returned after initial consolidation), no response, or pathologic fracture.

Results

The UBCs were located in the humerus ($n = 13$, 65 %), femur ($n = 6$, 30 %), and proximal fibula ($n = 1$, 5 %), with 5 (25 %) directly adjacent to the physis.

Uncomplicated cysts

At initial presentation only 2 of the 20 UBCs (one humeral, one femoral) were classified as uncomplicated. Uncomplicated UBCs displayed unilocular radiolucent

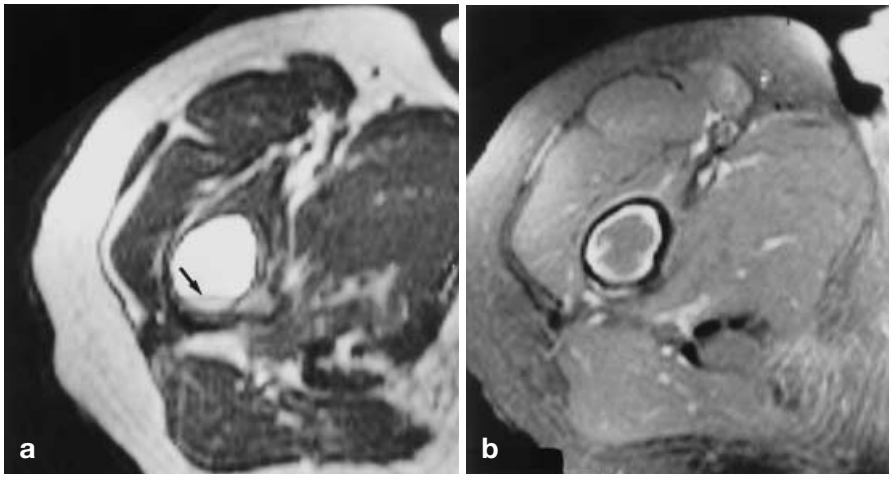

Fig. 1 a, b Uncomplicated unicameral bone cyst of the right proximal femur in a 4-year-old male. **a** T2-weighted MR scan shows prolonged relaxation time with homogeneous signal intensity of the contained fluid. A single fluid-fluid level is seen posteriorly (*arrow*). **b** After gadolinium infusion, the T1-weighted fat-suppressed MR image shows thin-rim peripheral enhancement

Fig. 2a–c Complicated unicameral bone cyst following acute pathologic fracture in a 12-year-old male. **a** Anteroposterior radiograph of the right proximal humerus reveals a multiloculated lytic lesion with pathologic fracture and "fallen-leaf" sign (*arrow*). **b** T1-weighted MR scan shows a low-intensity region with heterogeneous internal fluid signals, pathologic fracture, and "fallen-leaf" sign (*arrow*) inferiorly and a region of higher intensity superiorly, likely corresponding to hemorrhage (*arrow*). **c** T2-weighted MR scan shows a region with high signal intensity inferiorly, pathologic fracture, and 'fallen leaf' sign (*arrow*), and region of heterogeneous signal intensity superiorly (*arrow*)

features without evidence of periosteal reactive changes or ground-glass opacification on plain film. MR imaging showed fluid signals with homogeneous low and high intensities on T1- and T2-weighted images respectively (Fig. 1 a) and a fluid-fluid level in the femoral lesion ($n = 1$). Following gadolinium infusion, only thin-rim peripheral enhancement was seen (Fig. 1 b). Follow-up of these two UBCs indicated recurrence in both, with subsequent pathologic fracture in the humeral lesion.

Complicated cysts

The remaining 18 UBCs (90 %; 12 humeral, 5 femoral, 1 fibular) were complicated at initial presentation by either acute ($n = 4$, 22 %) or remote ($n = 14$, 78 %) pathologic fracture. Plain-film images taken at the time of MR imaging showed signs of healing with periosteal reaction ($n = 14$, 78 %) and ground-glass opacification ($n = 6$, 33 %). Intracystic septations were detected in 16 patients (89 %) by plain film.

On T1-weighted MR images, the fluid content of complicated UBCs displayed heterogeneous ($n = 9$, 50 %; Fig. 2) or homogeneous ($n = 9$, 50 %) signals. On T2-weighted images, the fluid signal was heterogeneous ($n = 15$, 83 %; Fig. 2) or homogeneous ($n = 3$, 17 %). MR scans detected septations in all these lesions and also found one or more fluid-fluid levels or internal hemorrhages ($n = 11$, 61 %; Fig. 2). In two cases multiple fluid-fluid levels were seen within a single lesion. MR imaging also showed adjacent soft-tissue changes ($n = 2$; Fig. 3) and "fallen-leaf" sign ($n = 1$; Fig. 2).

Fat-suppressed gadolinium-enhanced images were obtained in 15 (83 %) of the 18 complicated UBCs. All showed significant enhancement with focal (large nodules, $n = 5$; and small loci, $n = 4$), subcortical ($n = 2$, Fig. 3 a), thick peripheral ($n = 9$, Fig. 3 b), and heterogeneous ($n = 7$) patterns. However, all the complicated UBCs contained at least one focal region with homogeneous fluid signals and thin-rim or septal enhancement.

Follow-up for the complicated UBCs indicated complete healing ($n = 13$, 72 %; representing 75 %, 60 %, and 100 % of the complicated humeral, femoral, and fibular lesions, respectively), incomplete healing ($n = 2$), recurrence ($n = 2$), no response ($n = 1$), and refracture ($n = 2$, 11 %; 1 humeral and 1 femoral).

Correlation between plain film and MR images

For the six complicated UBCs exhibiting ground-glass opacification on plain film, the region of opacification corresponded to focal homogeneous enhancement

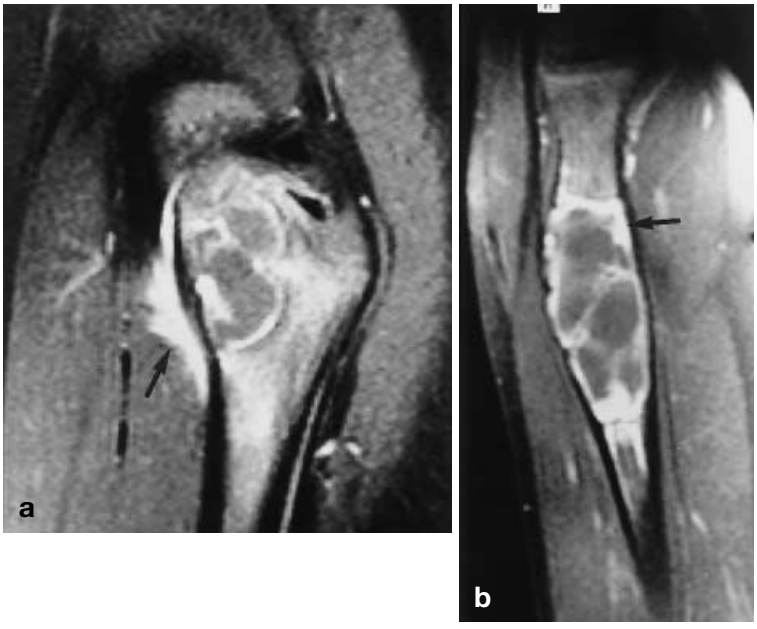

Fig. 3a, b Variable enhancement on T1-weighted fat-suppressed MR imaging in two children after gadolinium infusion. **a** Complicated unicameral bone cyst in the femur of a 1-year-old male showing subcortical and adjacent soft-tissue enhancement (*arrow*). **b** Complicated unicameral bone cyst in the humerus of an 8-year-old male, showing thick peripheral enhancement (*arrow*)

(large nodules, $n = 5$; Fig. 4) after gadolinium infusion. (No enhanced image was available in one case.) MR imaging also proved effective in detecting intracystic septations: these were identified in 16 (89%) complicated UBCs by plain film and in all by MR.

Histology

An intraoperative biopsy was performed on the fibular lesion. On MR imaging this lesion had a distal region with homogeneous fluid signals and thin-rim enhancement and a proximal region with heterogeneous signal intensities and enhancement (Fig. 5). Histologic examination showed UBC in the distal region, and aneurysmal bone cyst in the proximal region (Fig. 5).

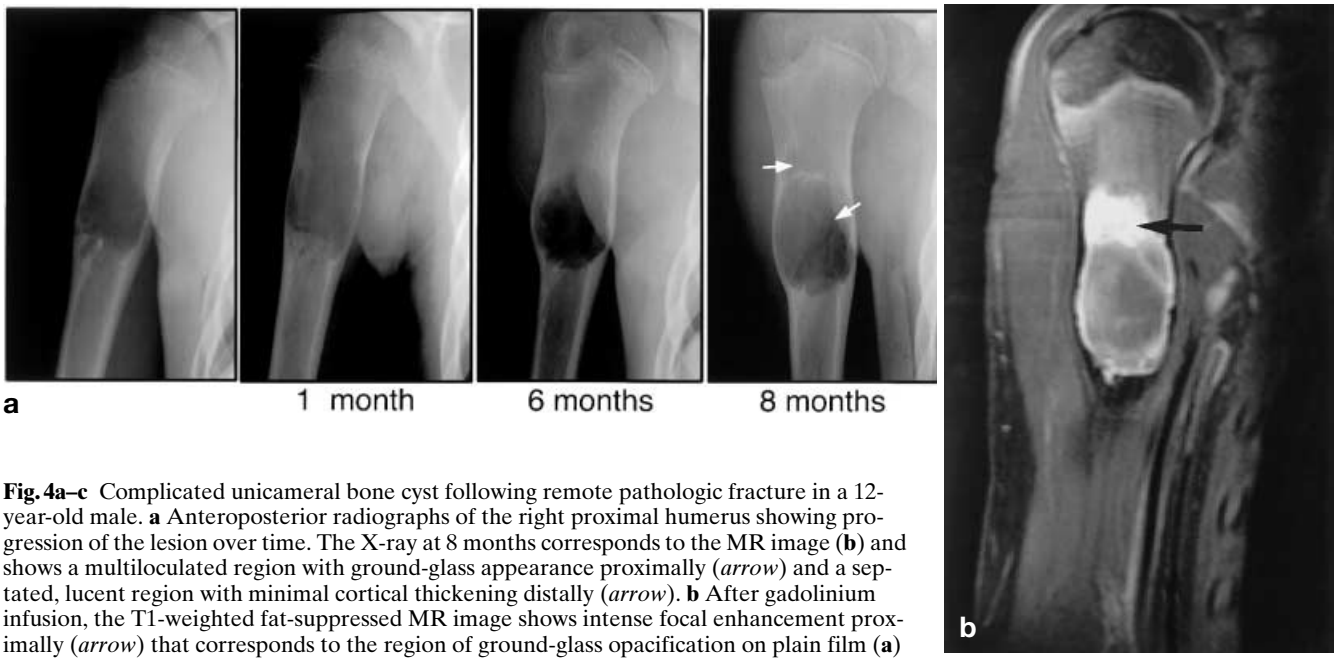

Fig. 4a–c Complicated unicameral bone cyst following remote pathologic fracture in a 12-year-old male. **a** Anteroposterior radiographs of the right proximal humerus showing progression of the lesion over time. The X-ray at 8 months corresponds to the MR image (**b**) and shows a multiloculated region with ground-glass appearance proximally (*arrow*) and a septated, lucent region with minimal cortical thickening distally (*arrow*). **b** After gadolinium infusion, the T1-weighted fat-suppressed MR image shows intense focal enhancement proximally (*arrow*) that corresponds to the region of ground-glass opacification on plain film (**a**)

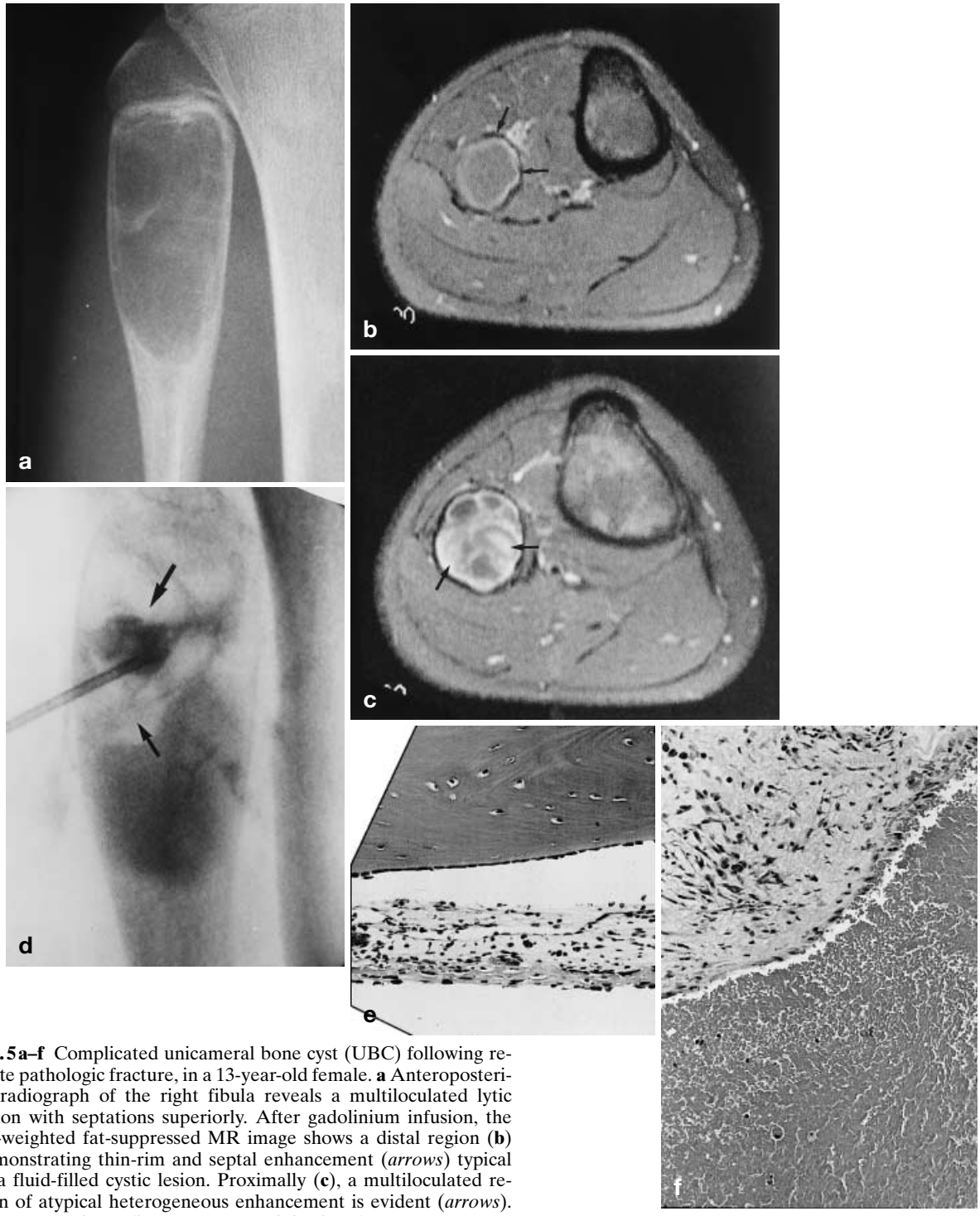

Fig. 5a–f Complicated unicameral bone cyst (UBC) following remote pathologic fracture, in a 13-year-old female. **a** Anteroposterior radiograph of the right fibula reveals a multiloculated lytic lesion with septations superiorly. After gadolinium infusion, the T1-weighted fat-suppressed MR image shows a distal region (**b**) demonstrating thin-rim and septal enhancement (*arrows*) typical of a fluid-filled cystic lesion. Proximally (**c**), a multiloculated region of atypical heterogeneous enhancement is evident (*arrows*). **d** Intraoperative radiopaque contrast injection shows poor contrast distribution proximally (*arrows*), demonstrating the multilocular nature of this region. **e** Photomicrograph of the distal region shows UBC with a thin cyst membrane (hematoxylin and eosin, × 40). **f** Photomicrograph of the proximal region shows aneurysmal bone cyst with a thick cyst membrane (hematoxylin and eosin, × 40)

Discussion

In our series, clinical histories were used to subdivide the UBCs into two groups: those with or without a history of acute or remote pathologic fracture. This classification was based solely on this factor [2] and is unrelated to other systems [7], in which UBCs are termed active (in the process of expanding) or inactive. Active UBCs are typically unilocular, adjacent to the physeal plate, and occur in younger patients [7].

MR imaging of the two uncomplicated UBCs showed homogeneous signal intensities with prolonged relaxation times (Fig. 1). These relaxation times are secondary to fluid within the cyst, a finding consistent with previous reports of UBC [1, 2–5] and typical of benign cystic lesions [4, 9]. However, most cysts in our series ($n = 18$, 90%) had undergone previous fracture and were classified as complicated at the time of imaging. This was an expected finding as UBCs usually present after pathologic fracture. The fluid component of complicated cysts displayed heterogeneous signal intensities on T1- and T2-weighted MR images (Fig. 2).

The heterogeneous MR fluid pattern of the UBC following pathologic fracture has been documented [1, 2], but not in a series as large as ours. Previous hemorrhage contributes to the variability of the cyst fluid, which can appear clear, yellow, orange, red or brown at the time of surgery [1]. An interesting finding was the high prevalence of fluid-fluid levels and internal hemorrhages in the complicated UBCs of our series. Moreover, in two cases multiple fluid-fluid levels were seen within the same lesion. Fluid levels are due to the settling of degraded blood products from previous hemorrhages and have been reported in isolated case reports of UBC [1, 6] and other tumors of bone and soft tissue [10]. Over time these blood products form calcified fibrin coagula, a common histologic finding in UBCs [11].

Further histologic changes occur in UBCs after pathologic fracture and may contribute to inhomogeneous cyst content. These changes include the formation of fibrous septa and newly formed bone trabeculae [1, 12]; all the complicated UBCs in our series demonstrated septations on MR imaging. Other histologic features after pathologic fracture include foci of granulation tissue and calcium deposits [1, 12]. Another common finding is acellular fibrin-like material, surrounded by osteoblasts, which resembles odontogenic cementum and appears dense on plain film [1, 13].

The focal, thick peripheral, heterogeneous, and subcortical enhancement patterns, as well as the adjacent soft-tissue changes seen in the complicated UBCs, have not been reported previously. While such MR findings are not suggestive of benign cysts [9], they have been reported in other cystic bone lesions [1]. The pathologic changes known to occur in UBCs after fracture and with subsequent early healing likely account for these enhancement patterns. Adjacent soft-tissue changes (Fig. 3a) are typical of healing with periosteal reaction [1]. Focal nodules of enhancement correlated with regions of ground-glass opacification observed on plain film (Fig. 4), a feature of consolidation, calcification, and new bone trabeculation [8].

The two uncomplicated UBCs demonstrated thin-rim enhancement (Fig. 1c); a finding documented previously [1] and likely due to the cyst's thin fibrovascular membrane [1, 7]. This enhancement contrasted with the thick peripheral enhancement (Fig. 3b) seen in the complicated UBCs of our series. The cyst membrane is known to become as thick as 1 cm following pathologic fracture as a result of reparative callus or reactive fibrosis [14]. This thickened membrane is also highly vascular and bleeds when touched by the surgeon; it likely accounts for the thick peripheral enhancement patterns.

Another significant finding in our series was that focal nodules of homogeneous enhancement on T1-weighted fat-suppressed gadolinium-enhanced MR imaging correlated with regions of ground-glass opacification on plain film ($n = 5$, Fig. 4). Ground-glass opacification has been shown to indicate cyst healing [8]. In our series, all regions of focal enhancement subsequently consolidated on follow-up. In these five cases, treatment methods varied: one patient received a steroid injection, three received bone-marrow injection, and one patient was not treated.

Enhanced MR images were highly effective in detecting intracystic septations, finding them in all complicated UBCs compared with 89% by plain film. Improved treatment outcomes are associated with fewer intralesional septa, as seen by cystogram, allowing for improved steroid distribution within the cyst [15].

In our series, the preliminary MR evaluation decreased operating room time by recognizing areas requiring selective intra-operative contrast injection. Although anecdotal, we found that fat-suppressed T1-weighted MR images helped localize residual cystic components present in complicated UBCs.

Additionally, our series demonstrated the lack of uniform response to therapy during clinical management of UBC. Despite the various treatment options available, failure of therapy has been documented in 10% to 20% of cases [1]. Responses to therapy may differ as a result of the variable internal characteristics of UBCs at presentation, a feature supported by our MR findings.

One complicated fibular UBC (Fig. 5) had regions of both thin-rim and heterogeneous enhancement. Histologic examination of the characteristically cystic region with thin-rim enhancement revealed UBC, while examination of the heterogeneous region indicated aneurysmal bone cyst (ABC). The association between UBC and ABC is not uncommon [1]. Moreover, the incom-

plete fibrous septa that form within a UBC after pathologic fracture are lined by fibroblasts and giant cells, creating an appearance that may be indistinguishable from an aneurysmal bone cyst [1, 12]. Our study, specifically the two cases with multiple fluid-fluid levels, further substantiates this association.

A limitation of our study is that, as a retrospective review, histologic evaluation was available only for lesions that appeared poorly demarcated at the time of surgery ($n = 2, 10\,\%$). UBCs rarely require biopsy and histologic analysis is not usually carried out. Therefore, few direct histologic data can be correlated with the MR images. Additionally, our study was not designed to demonstrate the value of MR imaging in diagnosing UBC. Instead we reviewed and characterized its MR appearance.

In conclusion, a comprehensive review of 20 lesions initially diagnosed and treated as UBCs demonstrated that these lesions frequently appear complicated on MR imaging because of acute or remote pathologic fracture and early healing. Heterogeneous fluid signals and regions of nodular and thick peripheral enhancement are likely related to previous pathologic fracture and early healing. Focal nodules of homogeneous enhancement correlated with ground-glass opacification on plain film, and consolidation on follow-up.

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