Extraosseous migration of a primary aneurysmal bone cyst after CT-guided core needle biopsy

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INTRODUCTION

Aneurysmal bone cyst (ABC) is a benign expansile tumor characterized by blood filled cyst-like cavities separated by connective tissue septa containing spindle cells, osteoclast type multinucleated giant cells, and reactive woven bone or osteoid.[1,2] Most ABCs arise de novo (primary ABCs).[1] Nonetheless, there also are ABCs (secondary) that are associated with another benign or malignant bone lesion.[1] ABC usually appears on radiographs as an eccentric, lytic and expansile lesion with a well-defined margin.[2] MRI may show a lesion bordered by a thin rim of low signal, an increased signal with augmented T2 weighting, and a lobulated contour with fluid–fluid levels.[2]

ABC typically occurs in the metaphysis of long bones, and the posterior elements of the vertebrae in children and young adults.[2] Parosteal or subperiosteal ABC is a rare subtype of ABC, accounting for 7–9.3% of all ABC cases.[3] It is characterized by its "attachment" to a completely normal part of a bone. Soft-tissue or extraosseous ABC is a recently recognized and even rarer pathologic entity with histologic features identical to the ABC of bone, except for its location.[2] There are only sporadic reports of patients with extraosseous ABCs in the English literature.[4] We present (to the best of our knowledge) the first case of extraosseous migration of a primary aneurysmal bone cyst after a CT-guided core needle biopsy. Informed consent for publication of medical information was obtained from the patient's parents.

CASE REPORT

A 14-year-old girl with a cystic lesion located at the proximal third of the left femur was referred to our clinic for further evaluation and treatment. The patient and her parents reported a brief history of gradually increasing dull pain at the left groin area with an onset approximately 2 months before her initial evaluation. Range of motion of the left hip joint was normal. Blood tests also were within normal range. Plain radiographs (Figure 1), an MRI (Figure 2) and a Tc^{99} bone scan (Figure 3) were consistent with an ABC. Our initial diagnosis was confirmed by a CT-guided core needle biopsy (Figure 4).

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The patient's surgery was scheduled for 1 week after the biopsy, and she was advised to avoid weight bearing on the affected limb during that time. Being completely uncooperative, she continued full weight bearing, until eventually she fell and sustained a pathologic fracture of the neck of the affected femur (Figure 5). The fracture was treated conservatively (skin-traction with the limb in abduction). Six weeks later, radiographs revealed the fracture to be completely healed. Nevertheless, movement of the affected hip joint was painful in all directions, and plain radiographs revealed the outline of a large mass originating from the proximal third of the femur (Figure 6). The mass was easily identifiable beneath the anterior thigh musculature and painful on palpation. White blood cell count (WBC), erythrocyte sedimentation rate (ESR), and C-reactive protein (CRP) evaluations all were within normal range. An MRI confirmed the existence of a large lobulated cystic mass adjacent to the proximal third of the femur. Because of the rapid progression of the lesion, a second biopsy was deemed necessary; it was performed under CT-guidance as well. The initial diagnosis of ABC was re-confirmed, and the patient once again was scheduled for surgery.

One day before the operation, the patient underwent a digital subtraction arteriography (DSA) followed by embolization of the lesion's main nutrient artery. With the patient under general anesthesia and through a typical anterior "Smith-Petersen" approach, the lesion was easily located and identified. However, the initial plan to completely excise the lesion altered intraoperatively because of the macroscopic appearance of the lesion and especially the soft and "rubber-like" texture of the lesion's anterior wall. An open biopsy was performed instead to ascertain the diagnosis. The third biopsy confirmed once again the initial diagnosis of ABC.

Five months after the initial evaluation of the patient, the radiographic appearance of the lesion was consistent with a typical extraosseous ABC, albeit attached with a small part of its posterior aspect to the anterior aspect of the proximal femur (Figure 7). The patient underwent (through the same anterior "Smith-Petersen" approach) a complete en-bloc excision of the cyst (Figure 8) and meticulous curettage of the remaining cavity in the proximal part of the femur, followed by the instillation of autologous bone graft. The pathologic examination of the completely excised extraosseous specimen confirmed once again the diagnosis of ABC. Gradually increasing weight bearing on crutches was allowed 1 month after the operation. Full weight bearing was achieved after another month. Two years later the lesion has clinically and radiographically completely resolved (Figure 9), with no signs of recurrence, and the patient currently is free of symptoms.

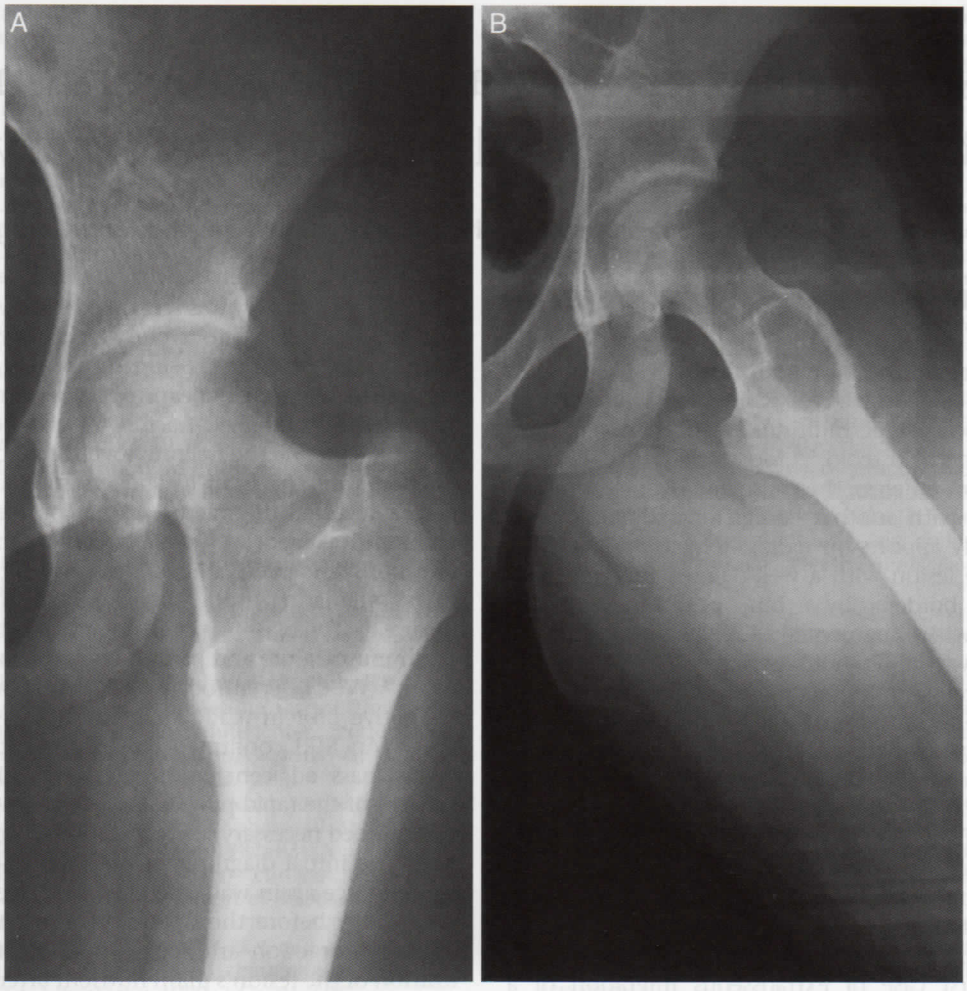

FIGURE 1. Anteroposterior (A) and lateral (B) radiographs of the affected hip depicting the typical eccentric, lytic and expansile characteristics of aneurysmal bone cyst.

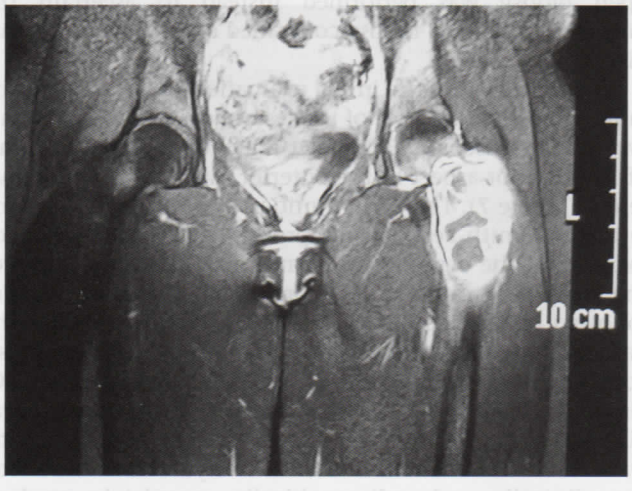

FIGURE 2. On MRI, the primary lesion appears to be multilobulated with bright signal on a T2-weighted image that is bordered by a thin rim of low signal.

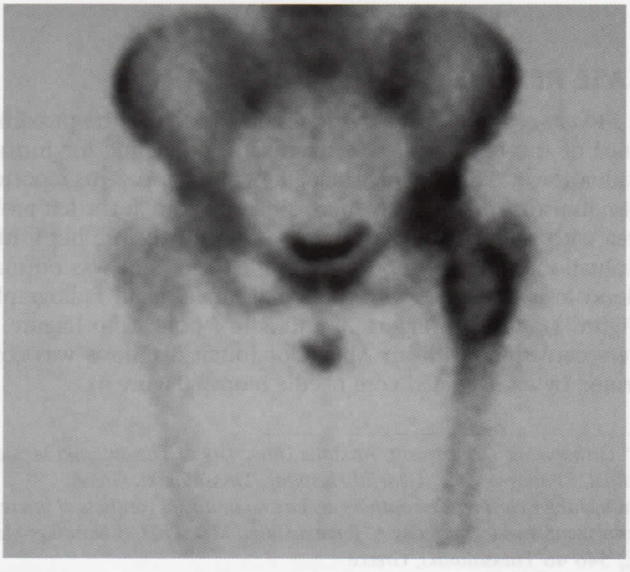

FIGURE 3. The Technetium99 bone scan with the typical "donut sign" representing increased uptake at the reactive border zone of the primary aneurysmal bone cyst.

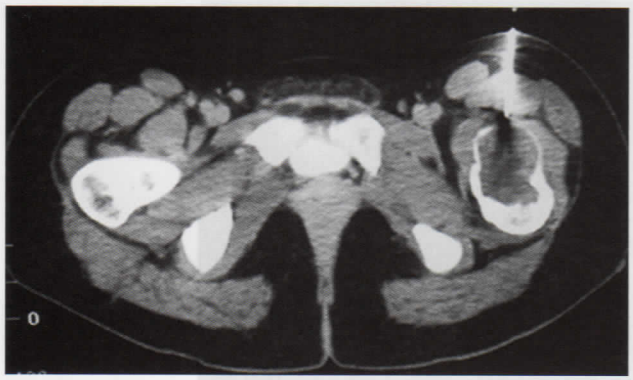

FIGURE 4. The CT-guided core needle biopsy of the primary lesion.

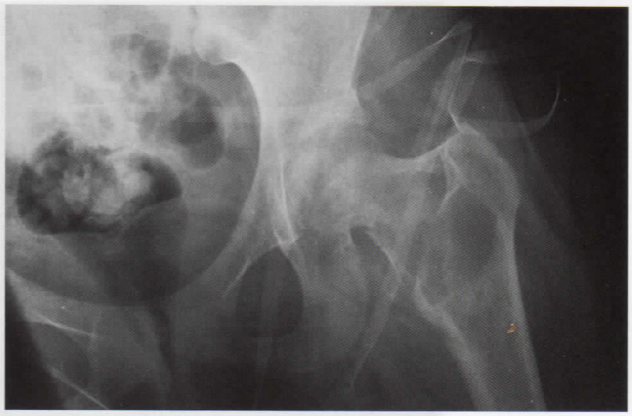

FIGURE 5. Anteroposterior radiograph of the affected hip showing the pathologic fracture at the femoral neck.

DISCUSSION

The soft-tissue counterpart of ABC, alternatively known as extraosseous ABC, is an extremely rare pathologic entity that may present with similar features on both imaging and histologic examination.[5] To the best of our knowledge, there are 20 reports of extraosseous ABCs in the English literature. Ranging in size from 2–9 cm, the reported tumors were located within the musculature of the upper[6–8] and lower extremities,[4,5,8–10] at the shoulder,[6,8] retroclavicular region,[11] hip,[12] groin,[8,13] pelvic cavity,[14] abdominal wall,[9] arterial wall of the bifurcation of the common carotid artery,[15] intracranial cavity,[1] larynx,[16] and hand.[17] None of these tumors showed any connection with adjacent bone.[4] Parosteal or subperiosteal ABC on the other hand certainly is more common than the extraosseous ABC but still a rare subtype of ABC.[3] Parosteal ABC is characterized by its "attachment" to a bone, which radiographically, macroscopically, and microscopically appears to be completely normal.

Our patient rapidly (in less than 2 months) developed a secondary extraosseous lesion after a CT-guided core needle biopsy that was performed to confirm the diagnosis of ABC located at the proximal part of the left femur. A possible explanation for the development of this secondary lesion is soft-tissue contamination from the biopsy procedure. This possibility is further enhanced by the fact that the first

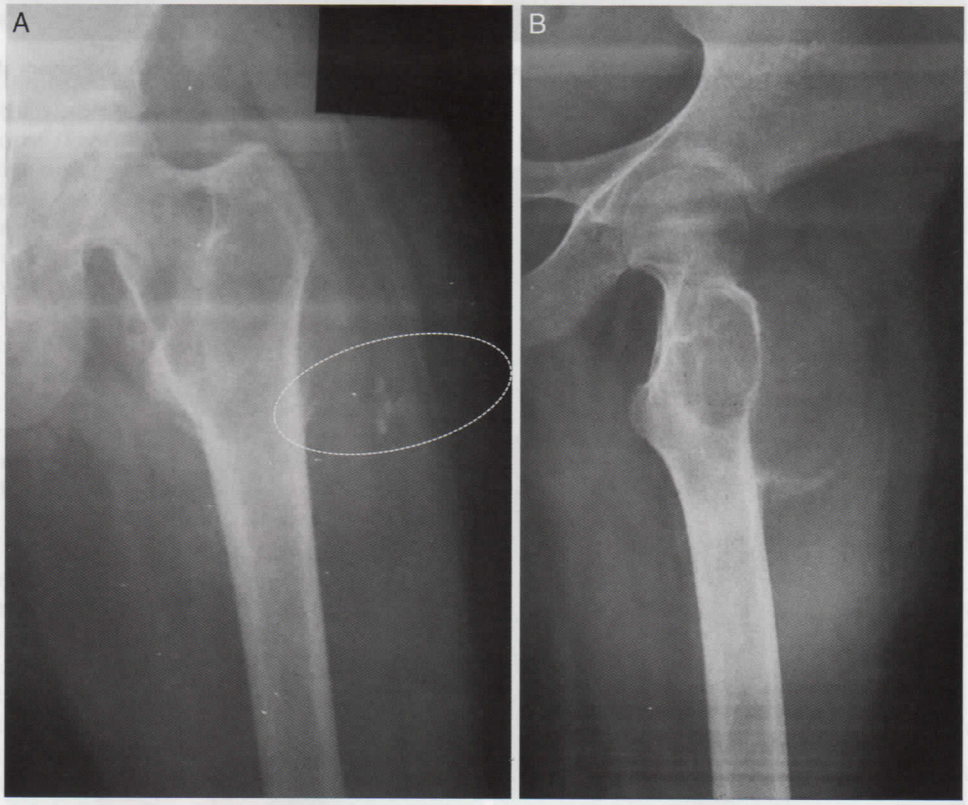

FIGURE 6. Anteroposterior (A) and lateral (B) radiographs of the affected hip after 6 weeks of conservative treatment for the pathologic fracture. Notice the outline of the secondary lesion originating from the proximal third of the femur and the first signs of primary mineralization of the lesion along the track of the biopsy (dotted line).

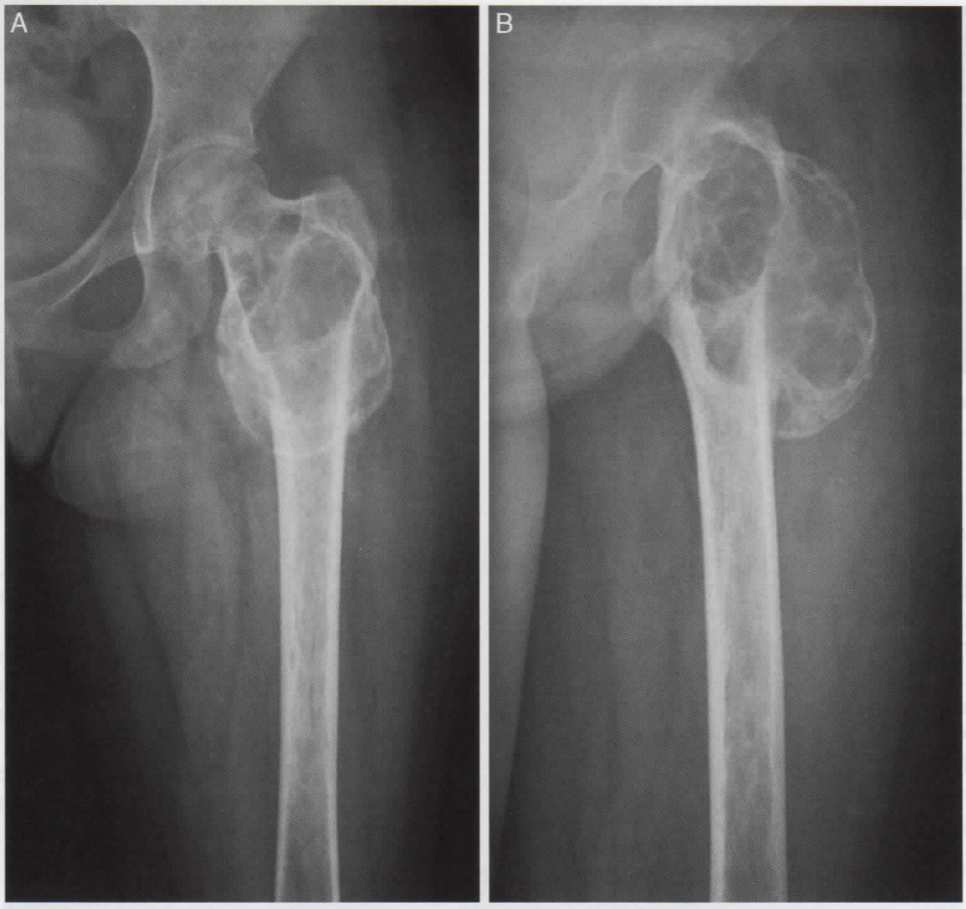

FIGURE 7. Anteroposterior (A) and lateral (B) radiographs of the affected hip 5 months after the initial evaluation of the patient showing the mineralized secondary aneurysmal bone cyst.

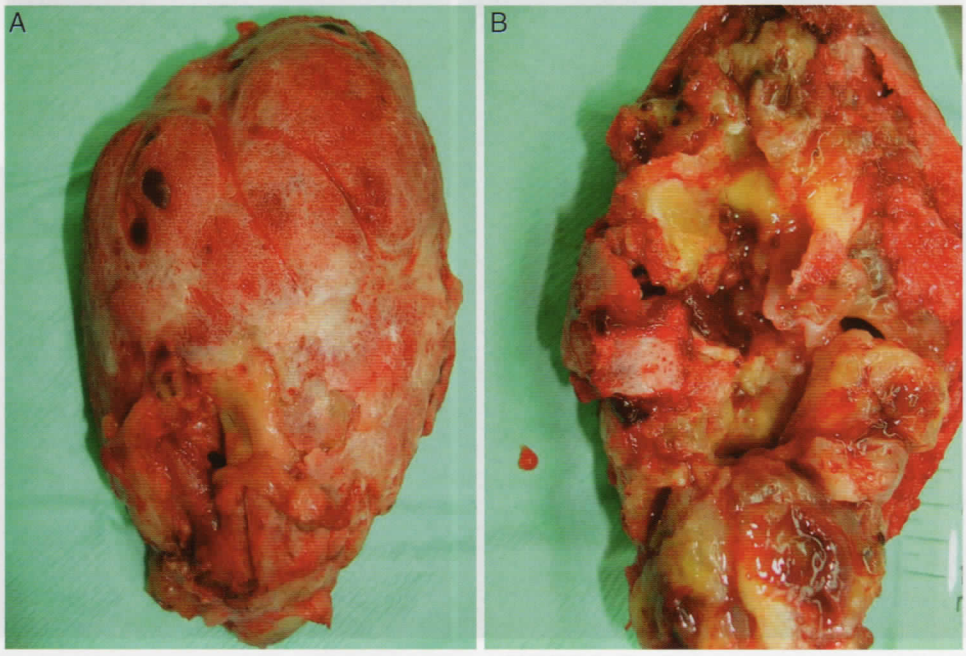

FIGURE 8. The anterior (A) and the posterior (B) aspects of the excised secondary aneurysmal bone cyst. Notice on the posterior aspect the parts with which the lesion was attached to the femur.

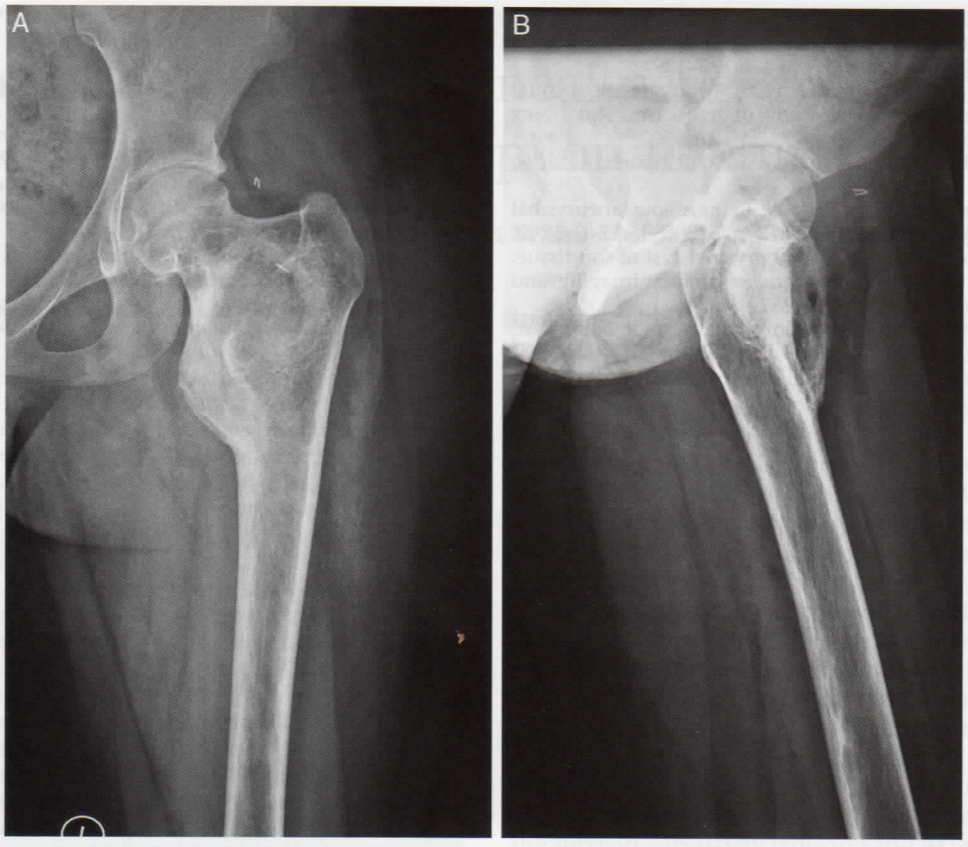

FIGURE 9. Anteroposterior (A) and lateral (B) plain radiographs of the affected hip 2 years after excision of the secondary aneurysmal bone cyst and the curettage of the primary aneurysmal bone cyst. The primary aneurysmal bone cyst has resolved and there are no signs of local recurrence of any lesion.

radiographically confirmed location of primary mineralization of the (until then) soft lesion could be identified along the track of the biopsy (Figure 6) as early as 8 weeks after the biopsy. Percutaneous needle biopsy is an accurate, rapid, safe and efficient procedure that often is used in the diagnosis of bone tumors. Nonetheless, there is always the theoretical and very low risk of recurrence of a malignant lesion in the track of the biopsy.[18] It is doubtful, however, if the possibility of soft-tissue contamination along the track of a biopsy really exists when a benign lesion is involved. Although it is less likely, the pathologic fracture (acting alone or combined with the biopsy) might have initiated the extraosseous migration of the primary ABC.

A CT-guided core needle biopsy, an open biopsy, and an excision biopsy all confirmed the diagnosis of ABC for this secondary lesion, which posed several diagnostic and therapeutic dilemmas. The secondary lesion looked radiographically like extraosseous ABC. Nevertheless, the lesion was attached to the femur from which it originated; hence, the diagnosis of extraosseous ABC had to be excluded. Although the secondary lesion had radiographic and macroscopic features consistent with parosteal ABC, we believe that this was not the case here, since there was one important difference (i.e. the existence of a primary lesion located within the femur adjacent to the extraosseous secondary lesion).

To the best of our knowledge this is the first report of secondary extraosseous development of ABC after a CT-guided core needle biopsy and a pathologic fracture. Even though one cannot be absolutely certain that the secondary lesion did not arise de novo, the rapid progression of the secondary lesion immediately after the biopsy and the fracture, the initiation of the lesion's mineralization procedure along the track of the biopsy, as well as the existence of important differences between this lesion and the parosteal and extraosseous subtypes of ABC, render extraosseous migration of the primary ABC lesion as a possible explanation for the development of its secondary counterpart.

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