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CASE REPORT

Uncommon observation of bifocal giant subchondral cysts in the hip: diagnostic role of CT arthrography and MRI, with pathological correlation

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Abstract Subchondral cysts (or geodes) are common in osteoarthritis (OA), usually in association with other typical signs, i.e., joint space narrowing, subchondral bone sclerosis, and osteophytosis. However, large lesions without the typical signs of OA or lesions located outside the weight-bearing areas are unusual and may be confused for other conditions, in particular, those of tumoral origin. We report the findings in a 48-year-old man who had been complaining of left buttock pain for 3 years, getting worse over the last year, and an evolutive limited range of motion of the hip. The pain was increased by weight-bearing and was not relieved by nonsteroidal anti-inflammatory drugs. Radiographs and CT showed a large multilocular lytic lesion within the femoral head and a large lytic lesion in the left ilio-ischiatic ramus, raising the question of bifocal tumoral involvement. On MRI, the lesions had low signal intensity on T1- and high signal intensity on T2-weighted MR images, with subtle peripheral enhancement on post-contrast T1-weighted images. CT arthrography, by demonstrating a communication between the femoral head and ischiatic cysts and the joint space allowed us to definitively rule out malignant conditions and to make the diagnosis of subchondral bone cysts. Total hip arthroplasty was performed. Pathological analysis of the resected femoral head and of material obtained at curettage of the ischiatic lesion confirmed the diagnosis of degenerative geodes. This case illustrates an atypical bifocal location of giant subchondral cysts in the hip joint mimicking lytic tumors, in the absence of osteoarthritis or rheumatoid arthritis, and highlights the role of CT arthrography in identifying this condition.

Keywords Hip · Tumors · Osteoarthritis · CT arthrography · MRI

Introduction

Subchondral cysts (also called geodes) are one of the major imaging radiographic features in osteoarthritis (OA), along with subchondral bone sclerosis and marginal osteophytosis. Most often, they are multiple and of limited size. Geodes are usually found in the weight-bearing areas: the femoral head and acetabular roof in the hip joint [1, 2]. The term "macrogeodes" has been proposed for lesions with a diameter superior to 1 cm [2]. Subchondral cysts greater than 2 cm are unusual [3].

Aside from OA, geodes in the femoral head and acetabulum may be observed in rheumatoid arthritis and ankylosing spondylitis [4, 5]. In the absence of OA or inflammatory coxitis, geodes may be found in osteochondritis dissecans, osteonecrosis, and hip dysmorphisms, such as coxa plana or polyepiphyseal dysplasia [6].

The exact pathogenic mechanism of the subchondral cysts remains under debate. Subchondral cysts observed alone without joint space narrowing and at a distance from weight-bearing areas may raise suspicion of a malignant process, increasing the need for further investigation [7].

This paper illustrates the case of a patient with bifocal "giant" cystic lesions located in the femoral head and in the

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posteroinferior part of the acetabulum, suggesting osteolytic tumors at initial imaging work-up.

Case presentation

A 48-year-old man was referred to our hospital, complaining of left buttock pain that had been evolving for 3 years, but had been getting worse during the last year. The pain was increased with weight-bearing and after physical exercise, with minor symptoms during the night, suggesting a mechanical origin. The pain required regular intake of paracetamol and nonsteroidal anti-inflammatory drugs. The patient, a construction worker, had decided to quit his job the year before.

His medical history was unremarkable apart from herniated lumbar disk surgery in 2009.

At physical examination, the patient had a limited range of motion of the hip: flexion 90°, extension 0°, abduction 20°, and adduction 10°. Internal and external rotations were painful and limited to 0° and 15° respectively. A limp was present, but the patient was able to walk without crutches.

Several weeks before referral, the general practitioner had requested an MRI of the lumbar spine because of the consistent pain. This study showed no spinal lesions.

Radiographs of the hips were performed (Fig. 1). Two lytic lesions were visible in both the left femoral head and the left ilio-ischiatic branch (Fig. 2). Comparison with the right hip showed shortening and broadening of the left femoral neck, and flattening of the left femoral head. CT confirmed the presence of lytic lesions in the left femoral head and ischium (Fig. 3). The question of possible bifocal tumoral involvement was raised. At the age of the patient, metastases or myeloma could be considered; primary tumors such as chondrosarcoma, giant cell tumor, etc., were less likely given the presence of two distinct lesions.

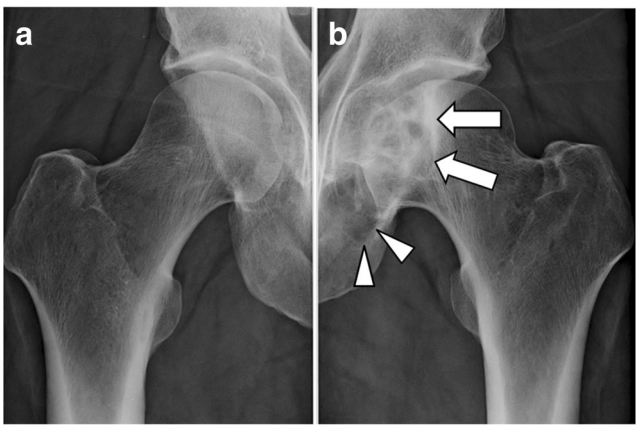

Fig. 1 Radiographs of the hips show **a** the normal appearance of the right proximal femur, and **b** osteolytic foci surrounded by sclerosis in the left femoral head (*arrows*) and ilio-ischiatic branch (*arrowheads*). Note dysmorphism of the left proximal femur, with a hypoplastic femoral head, and an enlarged and shortened femoral neck

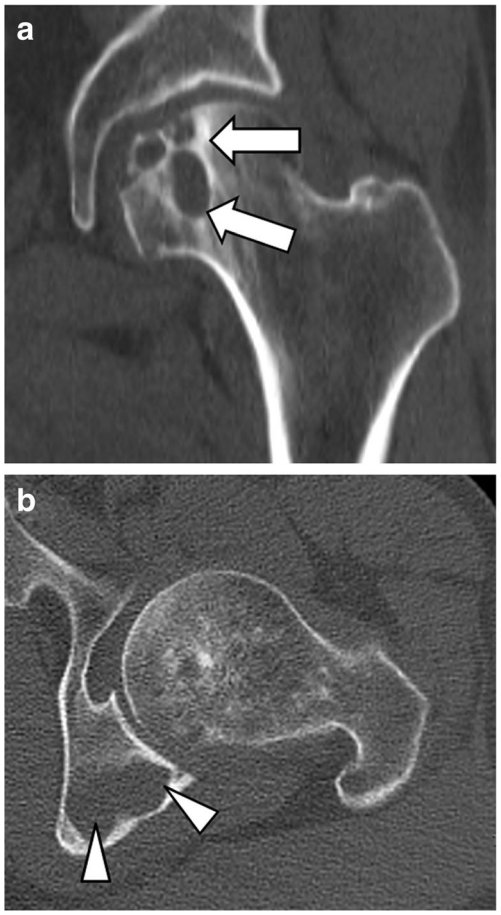

Fig. 2 Reformatted **a** coronal and **b** transverse. CT images show osteolytic lesions within the left femoral head (*arrows* in **a**), presenting a multilocular appearance and peripheral sclerosis, and a more homogeneous osteolytic appearance within the left ilio-ischiatic branch (*arrowheads* in **b**)

An MRI examination of the hip joint was obtained to characterize the lesions and rule out a tumoral process (Fig. 3). The examination revealed two large well-circumscribed lesions. One was located in the femoral head and the other in the posteroinferior part of the acetabulum. Their content was predominantly "hydrated," but more "solid parts" were present in the femoral head lesion, suggesting fibrous content (Fig. 3).

The main diagnostic hypothesis was that of giant subchondral cysts. But because the lesions were located in the non-weight-bearing areas of the hip, and because of the absence of typical signs of OA, i.e., subchondral bone sclerosis or marginal osteophytosis, a CT arthrography study was requested to evaluate the potential communication of the lesions with the joint. This examination demonstrated well-circumscribed lytic lesions, with penetration of contrast material originating from the joint space through small defects within the subchondral bone plate and partial opacification of the cyst's content (Fig. 4). Thus, this demonstration of the

articular origin of the lesions definitively confirmed the diagnosis of concurrent degenerative cysts (geodes).

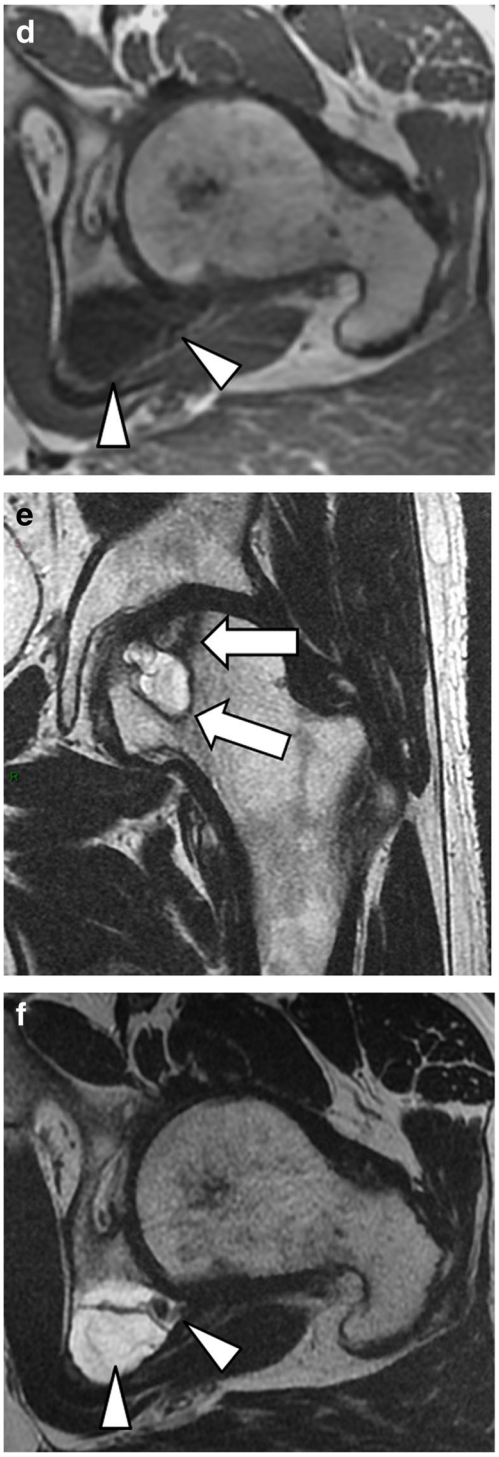

Fig. 3 Corresponding **a**, **b** coronal and transverse T1, **c**, **d** T1 after gadolinium injection, and **e**, **f** T2-weighted MR images show well-delineated foci of low signal intensity on the T1 and high signal intensity on the T2 images within the left femoral head (*arrows* in **a**, **c**, **e**) and ilio-ischiatic branch (*arrowheads* in **b**, **d**, **f**), with subtle peripheral enhancement

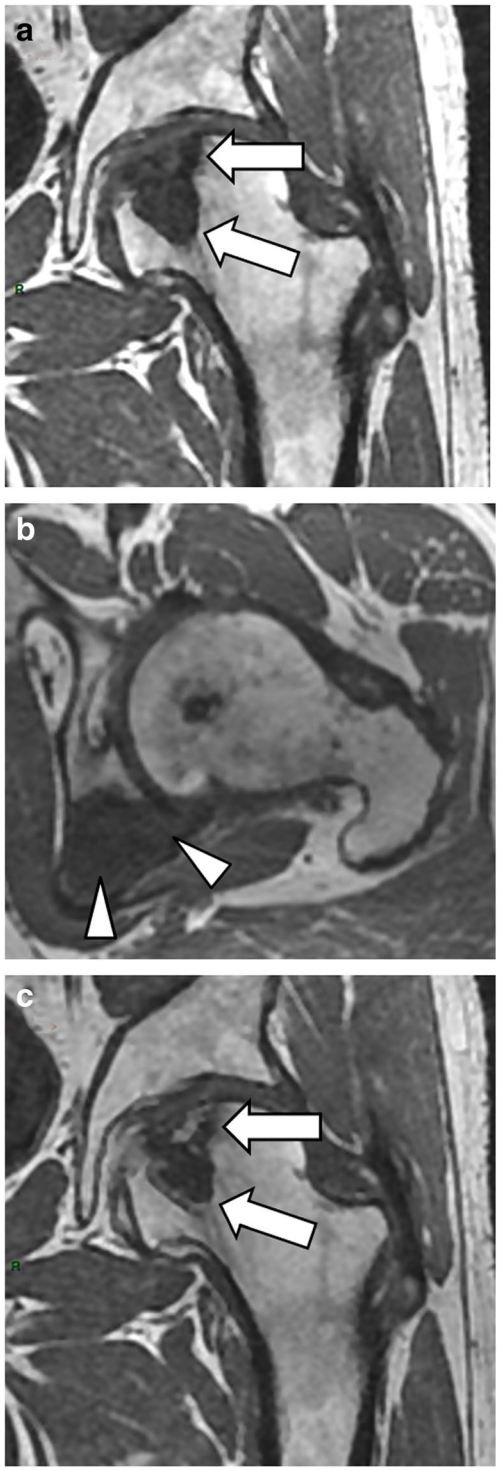

Fig. 3 continued.

A total hip arthroplasty was performed. After curettage, the ischiatic lesion was filled with bone auto-graft obtained from the resected femoral head. There were no complications and the patient could walk immediately on his left lower limb. He is completely asymptomatic at 9 months' follow-up.

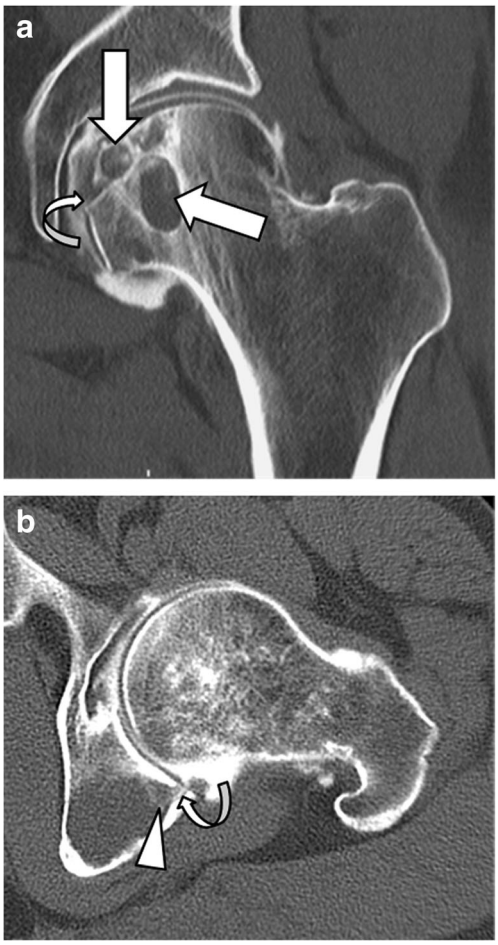

Fig. 4 Computed tomographic arthrography, reformatted **a** coronal and **b** transverse images show partial opacification of the lytic lesions of the femoral head (*arrows* in **a**) and the ilio-ischiatic tuberosity (*arrowhead* in **b**) by contrast material (compared with Fig. 2), and demonstrate the interruption of the subchondral bone plate, showing the articular origin of the cysts (*curved arrows*)

Pathological analysis confirmed the diagnosis of benign subchondral cysts. Analysis of the femoral head showed a mix of mucoid portions and more fibrous areas in a multiloculated cyst (Fig. 5). These areas were suggestive of more chronic portions of the cyst, with loss of communication with the joint and replacement of the mucoid content by fibrous tissue. There was also a subtle lymphoplasmocytic infiltration. The acetabular cyst exclusively contained viscous mucoid material of articular origin. No sign of malignancy was observed.

Discussion

Geodes in the femoral head are quite common [6, 8, 9]. Subchondral cystic lesions originating from the hip joint also commonly affect the acetabulum, sometimes with extension to the soft tissues [10, 11]. Large geodes are uncommon. Localization outside of the weight-bearing areas is also unusual. In the current case, the lesions were not only large and located in non-weight-bearing areas, but were also bifocal, suggesting a potential tumoral origin. To the best of our knowledge, this is the first report of such a concurrent femoral head and ischiatic giant subchondral cysts.

The incidence of geodes is variable in OA. In the SHOMRI study, geodes in the femoral head or acetabulum were found in 18% of patients [12]. Resnick et al. and Landells found geodes in the femoral head of all their patients [1, 2]. Rees et al. showed that acetabular geodes were present in 82 patients out of 100 suffering from hip osteoarthritis. All the cysts were located in the superior weight-bearing area of the acetabulum [13].

The incidence of "macrogeodes" has not been addressed specifically. Although hip OA is a frequent disease (affecting

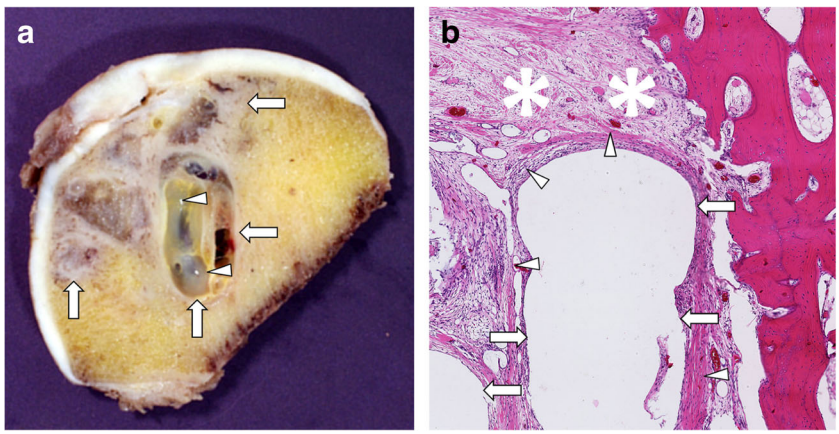

Fig. 5 Pathological correlation. **a** Macroscopic view of the resected femoral head (coronal slice) shows a multilocular cystic lesion of the femoral head (*arrows* in **a**). One of them contains mucoid material (*arrowheads* in **a**). **b** Microscopic view shows a mix of empty areas, corresponding to cystic components (empty of mucoid material owing to sample processing; *arrows* in **b**), surrounded by thin fibrous lining (*arrowheads* in **b**). Note the presence of loose connective tissue, presumably filling older cystic areas (*asterisk* in **b**) and surrounding bone sclerosis

5–10% of the population over the age of 55 years), solitary and large subchondral cysts are unusual. The observation of cysts or geodes in the absence of typical signs for OA and in a non-weight-bearing area may raise the suspicion of a neoplasm [3].

The exact origin of geodes is debated. Two theories exist regarding the pathogenesis of subchondral cysts, i.e., the "synovial breach theory" and the "bone contusion theory." The first theory suggests that synovial fluid might enter the subchondral bone through a breach in the cartilage and subchondral bone plate. This mechanism secondarily leads to bone resorption and cyst formation [2, 14]. The second theory suggests that repeated impacts onto the bony surfaces might cause fractures and vascular deficiency, which leads to subchondral bone necrosis and cyst formation [15]. Previous works have already demonstrated the communication path between large subchondral cysts in the femoral head and acetabulum and the joint space on CT scan [16]. The present observation supports this synovial breach theory as the CT arthrographic images clearly highlighted communications between the joint cavity and the subchondral bone, with small defects in the subchondral endplate, allowing the penetration of synovial fluid inside the bone. These communications are further demonstrated in our case by pathological correlation.

As some neoplastic conditions may be painful and mimic symptoms of hip OA, the diagnosis of a malignancy may be considered. Amongst potential lytic lesions observed in this area, chondroblastomas, giant cell tumors, clear cell chondrosarcomas or metastases could be considered [7].

This case highlights the role of CT arthrography to demonstrate the articular origin of the lesions. The technique confirmed the diagnosis of giant subchondral bone cysts and allowed a malignant origin to be ruled out, as the lesions were partially opacified and as the communications with the joint cavity were identified [17]. Interestingly, the same use has been made of CT arthrography, to demonstrate the articular origin of (extraosseous) cystic lesions observed in the soft tissues, with no evident communication between the lesion and joint with other techniques, especially MRI [18–21].

As Lequesne mentioned in 1985 and later in 1993, isolated giant geodes of the femoral head (that he called "macrogeodic cephalitis"), as observed in our patient, are often associated with, and seem to be the consequence of, chronic hip diseases such as osteochondritis dissecans during childhood or Legg–Perthes disease [6, 22]. The intensity of pain and discomfort remains moderate for years and the indication for surgery occurs later in adulthood. In the present case, the dysmorphic features of the proximal femur (neck shortening and widening, femoral head hypoplasia and flattening) were suggestive of minor sequelae of previous Legg–Perthes disease. The disease had been asymptomatic for years, with progressive expression during the last years evolving to very disabling symptoms.

Considering the hip joint damage, failure of the medical treatment, and the consistent pain and limited motion, our patient underwent total hip arthroplasty, which was considered to be the best therapeutic option for relieving the symptoms. There were few therapeutic alternatives. Percutaneous injection of cement within large cysts under CT and fluoroscopic guidance has been proposed as a rapid, safe, and efficient therapeutic option for symptomatic subchondral cysts, representing a minimally invasive alternative compared with classic surgery [23]. The present case report invites the cautious use of these percutaneous injections of cement in subchondral cysts because of the communications existing between the lesions and the joint cavity, suggesting a risk of intra-articular cement leakage with well-known harmful effects, such as chondrolysis [24].

In conclusion, in a patient with no typical signs of OA, an atypical observation of large concurrent subchondral bone cysts is reported, affecting the femoral head and ilio-ischiatic ramus, and mimicking tumoral foci on radiographs and CT. Facing this uncommon presentation, and to rule out an underlying malignant process, CT arthrography was used and proved helpful by demonstrating the articular origin of the lesions. The pathogenesis of subchondral cysts is still being discussed, and this case may support the "synovial breach theory."

Compliance with ethical standards

Conflicts of interest The authors declare that they have no conflicts of interest.

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