CASE REPORT

Multiple huge subchondral cysts associated with pseudogout in the bilateral knees: a case report and review of the literatures

Masaya Minoda · Tomoyuki Matsumoto · Seiji Kubo · Takehiko Matsushita · Koji Takayama · Yukiko Morinaga · Masahiro Kurosaka · Ryosuke Kuroda

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Introduction

Osteoarthritis (OA) presents as destruction and degeneration of articular cartilage, resulting in pathological changes to the subchondral bone [1]. Although the pathogenesis of bone changes is poorly understood, it appears to be influenced by alterations in abnormal mechanical forces around the affected joint. In addition to the degeneration of cartilage, characteristic radiological findings include narrowing of the joint space, subchondral sclerosis, and the appearance of osteophytes; intra-articular osteochondral bodies and subchondral cysts are all associated with OA.

Among these findings, subchondral cyst formation is often found mainly in OA and rheumatoid arthritis (RA) patients. Whereas RA is reported to cause huge synovial cysts [2–6], well known as geodes [7], huge subchondral cysts associated with pseudogout induced OA are extremely rare. We report an osteoarthritic patient with multiple huge subchondral cyst-like lesions in the bilateral knees who experienced several episodes of pseudogout attacks. Deep and large bone defects and severe lateral laxity due to multiple huge subchondral cystic lesions were successfully treated with semi-constrained type total knee arthroplasties with long stems and augmentation. No loosening was observed at the 1- and 2-year follow-ups.

A 79-year-old man presented at our hospital complaining of pain in the bilateral knees that had begun 2 years previously. Although there was no history of obvious injury, the patient had been diagnosed with pseudogout after several pain attacks, and calcium pyrophosphate dehydrate (CPPD) crystal deposits had been observed in the bilateral knees at an outpatient clinic. Upon visiting our institution, he complained of severe bilateral knee pain that had gradually worsened until he could no longer walk at any speed because of pain. On physical examination of the bilateral knees, the patient had a severely restricted range of motion with medial joint line tenderness, pain at extension and high flexion, and starting pain. There was slight swelling but no palpable joint effusion, redness, or local heat. The patient’s gait was slow, he used a walking stick, and exhibited a lateral thrust. Manual stress tests indicated severe lateral laxity with no anteroposterior instability. Neurovascular examination returned normal findings. Laboratory testing showed no apparent findings of infection, inflammation, or RA (CRP 0.3 mg/dl, ESR 8 mm/h, RF <12 IU/ml). Plain radiographs showed varus type osteoarthritic change in the form of a severely destructed femur and tibia, especially on the medial side, with marked osteophytes and several large cystic lesions (Fig. 1). Computerized tomography (CT) of both knees revealed huge multiple subchondral cystic lesions on the femur and tibia (Fig. 2). Magnetic resonance imaging (MRI) revealed the same intensity area as liquid in T2-weighted image (T2WI) and a high intensity area in T1-weighted fat suppression images (T1FS), indicating multiple cyst formation (Fig. 3, upper and middle panel). Another MRI slice revealed a fusion of cystic lesions and lipoma-like lesions showing isointensity with fat tissue with T2WI and T1FS (Fig. 3, lower panel).
The patient underwent total knee arthroplasty (TKA) of the bilateral knees, first of the right knee in May 2008 and then of the left knee in May 2009. On examining the medial compartment, extremely deep and large cyst formation was seen in the bilateral knees. Following bone resection of the femur and tibia, due to large bone defects and remarkable lateral instability, we applied a semi-constrained polyethylene insert and implants with long stems and metal augmentation (NexGen LCK) (Fig. 4), resulting in complete relief of the patient's symptoms. The pathology of cystic lesions showed fat tissue in most of the lesion and cysts with a wall of fibrous connective tissue, indicating a combination of subchondral cysts and lipoma-like lesion (Fig. 5, upper panel). In addition, the pathology of the synovium showed a moderate number and size of mononuclear inflammatory cells along lining cells and a moderate number of blood vessels, not suspicious for RA or its related diseases (Fig. 5, lower panel). One and 2 years after the operation in each knee, the patient has no symptoms related to the bilateral knees and was maintaining his daily activities. During this time, there were no symptoms and no laboratory data suspicious for RA or its related diseases.

The patient was informed that data from the case would be submitted for publication and gave his consent.
Discussion

Multiple huge subchondral cysts are often found in RA patients, and these were first reported as geodes by Jayson et al. [7]. Shih et al. [8] reported a huge tibial subchondral cyst successfully treated with standard stem and autologous bone grafting. However, there have been no reported cases of multiple huge subchondral cysts in osteoarthritic patients to our knowledge. In the present osteoarthritic patient, severe bone defects and lateral laxity due to multiple and huge cystic lesions compelled us to use a semi-constrained type prosthesis with a long stem and metal augmentation. The treatment provided the patient with relief of pain and stable daily life. However, the follow-up period is as short as 2 years; thus, a longer term follow-up is needed to investigate the real clinical usefulness of the treatment.
Generally, there are two main theories that attempt to explain the development of osteoarthritic cysts. The first is based on the idea that synovial fluid intrudes through the articular cartilage, resulting in hydraulic destruction of subchondral bone [9]. This theory is supported by the presence of defects in the articular cartilage resulting from cysts, of fragments of articular cartilage within cysts, and the similarity of cyst fluid to synovial fluid [10]. The second theory suggests that a localized area of subchondral necrosis of bone results from repetitive microtrauma, which leads to cystic degeneration in the bone while the articular cartilage is left intact [11, 12]. This is based on evidence of bony contusion, trabecular fracture, and primary subchondral osteolysis, which may subsequently communicate with the joint if the overlying articular cartilage and subchondral bone plate crack [13]. In our case, the existence of defects in the articular cartilage above the cysts partly indicates the validity of the first theory. However, as shown in the MRI finding in the lower panel of Fig. 3 and the histological finding in the upper panel of Fig. 5, it is of note that the cystic lesion seems to fuse into a lipoma-like lesion. Spjut et al. [14] reported a coalescence of smaller degenerative cysts as a mechanism for huge cyst formation. In addition, Wada and Lambert [15] reported the deposition of intraosseous fat in a degenerating simple bone cyst, resulting in a combination of subchondral cysts and lipoma-like lesion (upper panel; right: loupe, left: ×20). The pathology of synovium showing a moderate number and size of mononuclear inflammatory cells along lining cells and a moderate number of blood vessels, not suspicious for rheumatoid arthritis or its related diseases (lower panel; right: ×20, left: ×100).
indicating that the intraosseous cavity is partly filled with fat after involution of the cyst. Taken together, these previous reports and our findings suggest one degenerative subchondral cyst may fuse with another old intraosseous cavity filled with fat tissue. Accordingly, the coalescence of two different cysts at different stages may result in huge cyst formation.

In the present case, the patient was initially diagnosed with pseudogout based on the findings of severe pain and CPPD crystal deposits. In addition, histological findings (Fig. 5, lower panel) showed no evidence of RA or its related diseases. Cysts associated with CPPD are similar to those of degenerative joint disease in that they are commonly associated with joint space loss, eburnation, and a sclerotic rim. They may be numerous and larger than those associated with degenerative joint disease with fragmentation and collapse of the subchondral bone [16]. In the present case, the existence of pseudogout may have accelerated the degenerative change and huge cyst formation.

In conclusion, we reported a rare case of multiple huge subchondral cysts successfully treated with surgery. Deep and large bone defects and severe lateral laxity due to multiple huge subchondral cystic lesions were treated with semi-constrained type total knee arthroplasty with a long stem and augmentation, providing pain relief and patient satisfaction at the 1- and 2-year follow-ups.

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