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CASE REPORT

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Two cases of osteochondromatosis which developed in the iliopectineal bursa of an osteoarthritic hip

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Abstract Two osteoarthritis patients had osteochondromatosis in the iliopectineal bursa which communicated with the hip joint space. They received surgical resection of the cystic mass and total hip arthroplasty and had good clinical outcomes. The authors consider that these patients' osteochondromatosis was a secondary development on the synovium of the iliopectineal bursa due to chronic inflammation caused by osteoarthritis.

Key words Hip · Iliopectineal bursa · Osteoarthritis · Osteochondromatosis

Introduction

The iliopectineal bursa is a well-demarcated anatomic structure formed in the ilioinguinal region which can become inflamed in various hip diseases, e.g., osteoarthritis, rheumatoid arthritis, and pigmented villonodular synovitis. However, synovial osteochondromatosis rarely originates from the iliopectineal bursa in osteoarthritis patients. We examined two osteoarthritis patients who had osteochondromatosis originating from a large iliopectineal bursa who were successfully treated with bursectomy, or bursectomy with total hip arthroplasty (THA). We report their treatment courses and discuss the pathogenesis of secondary synovial osteochondromatosis in the iliopectineal bursa.

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Case reports

Patient no. 1 was a 79-year-old woman who had had left hip pain and gait disturbance for about 3 years. On the first examination, we found a tumor-like, soft and palpable mass, approximately 8 cm × 5 cm, on the lateral side of the left inguinal region. Radiographically, there was a bone cyst in the left femoral head and remarkable narrowing of the left hip joint space. The patient was evaluated as having end-stage osteoarthritis of the hip. Computed tomography (CT) and magnetic resonance imaging (MRI) also showed a cystic mass on the anterolateral part of the left hip joint, and this mass was partly connected to the joint capsule (Fig. 1). The patient underwent bursectomy with THA. The resected mass contained serous fluid and milky-white loose bodies (Fig. 2a). Histologically, some cartilaginous foci and bone formation were observed on the capsule of the cystic mass and loose bodies (Fig. 2b). Two years and 6 months have passed since the surgery, and the patient has no pain, she can walk without assistance, and there is no recurrence of the mass.

Patient no. 2 was an 80-year-old man. He detected a mass in the right inguinal region and visited this hospital. On the first examination, we found a palpable, soft, elastic, tumor-like mass, approximately 7 cm × 6 cm, in the right inguinal region. Radiographs revealed the presence of early stage osteoarthritis of the hip. MRI showed that the mass was continuous from the anterior part of the joint capsule. Because the patient did not have pain in the right hip joint, the mass was surgically resected but THA was not performed. No loose bodies were found within the cystic mass, but osteocytes were found on the capsule of the mass. Based on these findings, the patient was diagnosed as having synovial osteochondromatosis. Three years have passed since the surgery, and there has been no recurrence of the mass, and no progression of the osteoarthritis of the right hip.

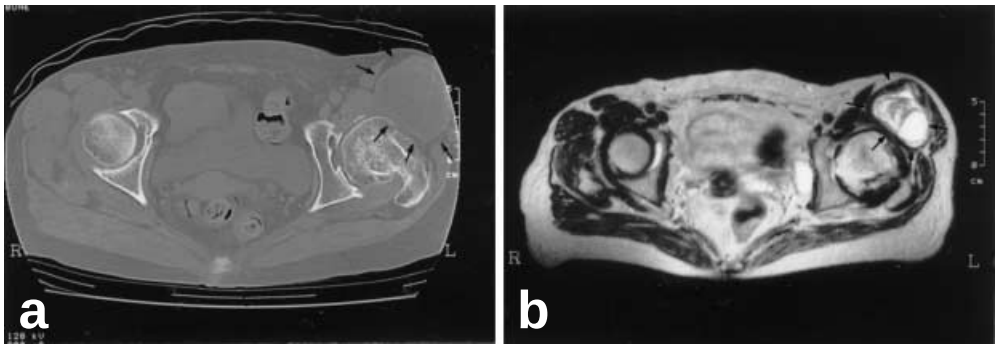

Fig. 1. Patient no. 1. Computed tomography scan (**a**) and magnetic resonance imaging (**b**) at hospital admission. Both show the cystic mass (*arrows*), approximately 8 cm × 5 cm, which partly communicated with the joint capsule on the anterolateral part

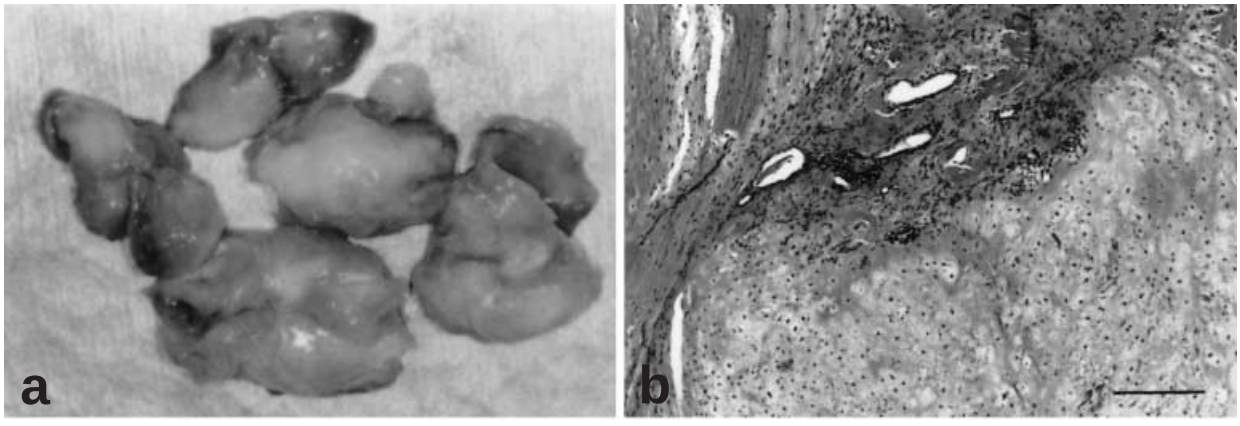

Fig. 2. Patient no. 1. **a** The cystic mass, which was surgically resected, contained serous fluid and milky-white loose bodies. **b** Histologically, some cartilaginous foci and bone formation were observed on the capsule of the cystic mass and loose bodies. HE staining. *Bar* 0.5 mm

Discussion

The iliopectineal bursa is the largest bursa in the human body, and is clearly demarcated by the iliac muscle on the anterior part and the iliopectineal eminence and joint capsule on the posterior part. Its medial edge reaches the iliopectineal eminence, and the lateral edge reaches the anterior inferior iliac spine. The average size is 6 cm × 3 cm, and approximately 15% of the human population are thought to have their iliopectineal bursa communicating with the hip joint space. The iliopectineal bursa is usually formed in the fetus, but it could be missed on rare occasions.[1]

The first known case of iliopectineal bursitis was reported by Fricke[2] in 1834 as an inflammatory disorder within the iliopectineal bursa. In 1933, O'Connor[3] conducted an anatomic, etiologic, diagnostic, and clinical study on 33 patients with iliopectineal bursitis and reported that its frequency was slightly higher in males than in females, age was not a factor, and possible causative factors are aggressive sports activity and heavy work in young people, and spur formation that could be caused by hip joint deformation in elderly people.

Communication between the iliopectineal bursa and the hip joint space occurs at a significantly high frequency in patients with hip joint diseases and this could cause coxitis. Therefore, several cases of iliopectineal bursitis complicated by hip joint diseases such as rheumatoid arthritis, osteoarthritis, and pigmented villonodular synovitis have been reported.[4,5] However, these two cases had the extremely rare condition of synovial osteochondromatosis within the iliopectineal bursa.

The causes of iliopectineal bursitis are still unknown, and several hypotheses have been proposed. Chandler[1] suggested that the communication between the hip joint space and the bursa is not formed by the growth of the iliopectineal bursa, but is attributable to chronic friction stimuli caused by the iliopsoas muscle. Binek and Levinsohn[6] suggested that the increased pressure in the joint space due to coxitis leads to the influx of the fluid in the hip joint space into the bursa, and this results in a large tumor-like mass. O'Connor[3] reported that regressive changes in the synovial membrane in the iliopectineal bursa could produce large cartilagenous foci or loose bodies.

Both of our cases had a clear communication between the hip joint capsule and the iliopectineal bursa. We therefore consider that (i) joint fluid which was overproduced within the hip joint capsule would flow into the bursa, and would be kept there by valve-like mechanism in the communication channel, resulting in a large tumor-like mass, and (ii) chronically repeated stimuli associated with

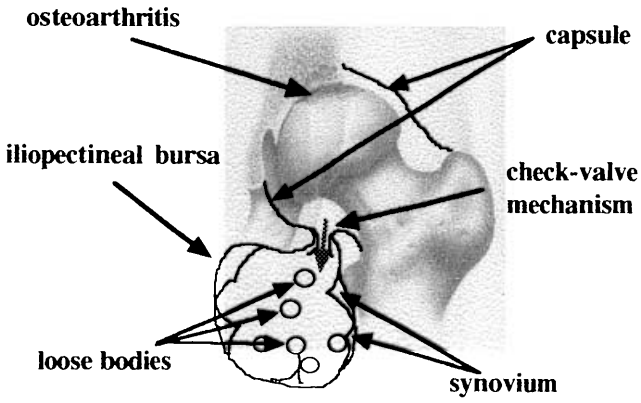

Fig. 3. The mechanism of generation of secondary synovial osteochondromatosis within the iliopectineal bursa. Osteochondromatosis in our patients was thought to have been generated by (i) the check-valve mechanism between the hip joint space and the iliopectineal bursa, which keeps the fluid from the joint space in the bursa, and (ii) chronic inflammation due to osteoarthritis

osteoarthritis would induce osteochondromatosis in the degenerated synovium of the bursa (Fig. 3).

Milgram[7] classified synovial osteochondromatosis into three stages based on clinical symptoms and histological findings, i.e., (1) there are active lesions in the synovium, but no loose bodies, (2) there are both active lesions in the synovium and loose bodies, and (3) there are no active lesions in the synovium, but there are many loose bodies. Patient no. 1 could be at stage 2, and patient no. 2 could be at stage 1.

Iliopectineal bursitis should be differentiated from inguinal hernia, lymphoma, iliopsoas abscess, femoral aneurysm, and tumors, and its differential diagnosis can sometimes be difficult. Generally, MRI and arthrography can provide accurate preoperative information. In our cases, osteochondromatosis in the iliopectineal bursa communicating with the hip joint space was diagnosed using MRI and CT.

The treatment for this disease is rest, followed by aspiration of the cystic content, and steroid injections. However, large cysts containing many loose bodies, or repeatedly recurring cysts, should be treated by incision or surgical resection. Malamed et al.[8] recommended THA in addition to surgical resection of the cyst. Their proposal would be reasonable for patient no. 1, who had serious associated osteoarthritis, in order to prevent a recurrence. Patient no. 1 underwent both resection of the cystic tumor and THA, and her clinical outcome has been good. We consider that her osteochondromatosis was generated in the synovium of the iliopectineal bursa because of chronic inflammation due to osteoarthritis.

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