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CASE REPORT

Femoral nerve palsy caused by a huge iliopectineal synovitis extending to the iliac fossa in a rheumatoid arthritis case

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Abstract We report on a 54-year-old woman with rheumatoid arthritis who had severe femoral nerve palsy affected by a distended synovium in the hip joint. Surgical exploration demonstrated a perforation of the iliopectineal bursa connecting with the hip joint. The patient fully recovered from femoral nerve palsy after surgery. It was considered that synovitis of the hip joint had developed following huge iliopectineal bursitis.

Keywords Femoral nerve palsy · Hip joint · Iliopectineal bursa · Rheumatoid arthritis · Synovitis

Introduction

Iliopectineal bursitis has been reported to develop subsequent to various diseases such as rheumatoid arthritis (RA) [1–4], osteoarthritis [5], total hip replacement [6], pigmented villonodular synovitis [7], and hemodialysis [8]. However, it is rare for synovitis to cause the bursa to enlarge sufficiently to cause severe femoral nerve palsy. We report the course of treatment and discuss the pathogenesis of an enlarged bursa with synovitis.

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Case report

A 54-year-old woman was diagnosed with RA at the age of 52, and was treated by drug therapy (prednisolone 5 mg/day, lobenzarit disodium 240 mg/day, bucillamine 300 mg/day) at a previous hospital. She was diagnosed as having destruction of the right hip joint at that hospital 2 weeks after she experienced right thigh pain and multiarticular pain. She was referred to Tsukuba University Hospital, and we detected a mass in the right inguinal region accompanied by femoral nerve palsy.

A hard elastic, mass was palpable in her right inguinal region with fluctuation. Marked hypesthesia and numbness were observed in the femoral nerve region from the anterior front of the thigh to the medial malleous of the tibia. The patient could not extend her right hip joint because of right thigh pain. Her right leg was shortening. The circumference of the right thigh demonstrated atrophy compared with that of the left thigh, and she could not extend her knee against gravity. The right patellar tendon reflex was absent.

Hematological examination demonstrated a WBC count of 9,900 μl^{-1}, C-reactive protein (CRP) of 14.69 μg/dl, erythrocyte sedimentation rate (ESR) of 130 mm/h, matrix metalloproteinase-3 (MMP3) of 334.4 ng/ml, and rheumatoid factor (RF) of 50 IU/ml.

X-ray examination demonstrated destruction of the right femoral head and an enhanced soft tissue shadow around the right hip joint (Fig. 1). The destruction of the femoral head and cortical atrophy of the femur were more progressive compared with assessment at the previous hospital

Fig. 1 Plain XP at the time of hospitalization. The right femoral head was destroyed, and the soft tissue shadow was enhanced around the right hip joint

within a short period. Computed tomography (CT) demonstrated an encapsulated cystic mass adjacent to the iliac bone. The capsule was enhanced with contrast medium, and the contents of the capsule were identified as fluid. The cyst was connected with the hip joint (Fig. 2a) and was dumbbell-shaped, forming a bridge over the iliac bone (Fig. 2b). The femoral vein was compressed to flatness and the volume of the cyst calculated via three-dimensional CT was 480 ml (Fig. 3). Magnetic resonance imaging demonstrated that the cystic fluid was serous.

Electrophysiological study was performed. We could not induce a muscle response of the quadriceps femoris, and the electromyogram demonstrated a denervation pattern of the femoral nerve.

A percutaneous puncture was performed to confirm the diagnosis. The fluid was viscous and orange in color, and the volume was 75 ml. There was no bacterial organism identified on either culture or Gram stain.

Surgical excision was carried out via an anterior approach. The cyst was located under the iliacus muscle, which was shifted medially, and the femoral nerve was compressed upward under the inguinal ligament (Fig. 4). The cyst was connected to the capsule of the hip joint, and part of the capsule was extracted. We resected the inguinal ligament to approach the area under the iliacus muscle. The cyst was resected piece by piece because the membrane of the cyst was very thin and adhered to the surrounding soft tissue. The cystic fluid was the same as previously obtained by puncture, and there was a considerable quantity of a pale yellow substance in the cyst and hip joint. After resection, the tension on the femoral nerve was decreased. Histological examination demonstrated synovial proliferation with a fibrinoid necrotic

Fig. 2 **a** Cyst CT at the axial hip joint level. The capsule was enhanced with contrast medium and its contents were identified as fluid. The cyst was connected to the hip joint. **b** Sagittal CT. The cyst was dumbbell-shaped, forming a bridge over the iliac bone

tissue that was consistent with typical RA. Because the hole in the capsule was big, we could not suture it and so covered it with the iliacus muscle and carefully sutured the area.

The patient could contract her right quadriceps voluntarily at 4 weeks after the first surgery. Because infection was ruled out, chemical therapy relieved multiarticular pain, and muscle tonus increased sufficiently to prevent hip joint dislocation, we performed total hip arthroplasty through a posterolateral approach 6 weeks following the first surgery. The hole in the capsule had disappeared and the capsule appeared intact on the second surgery. The quadriceps had fully recovered 4 months after the first surgery, the patient could walk normally and had no

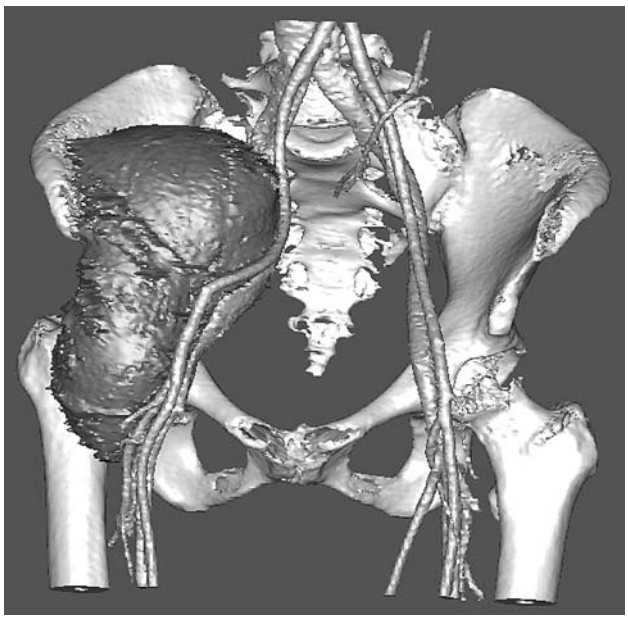

Fig. 3 Three-dimensional CT demonstrated a dumbbell-shaped cyst over the iliac bone, and the femoral vein was flattened due to compression. The volume of the cyst calculated from three-dimensional CT was 480 ml

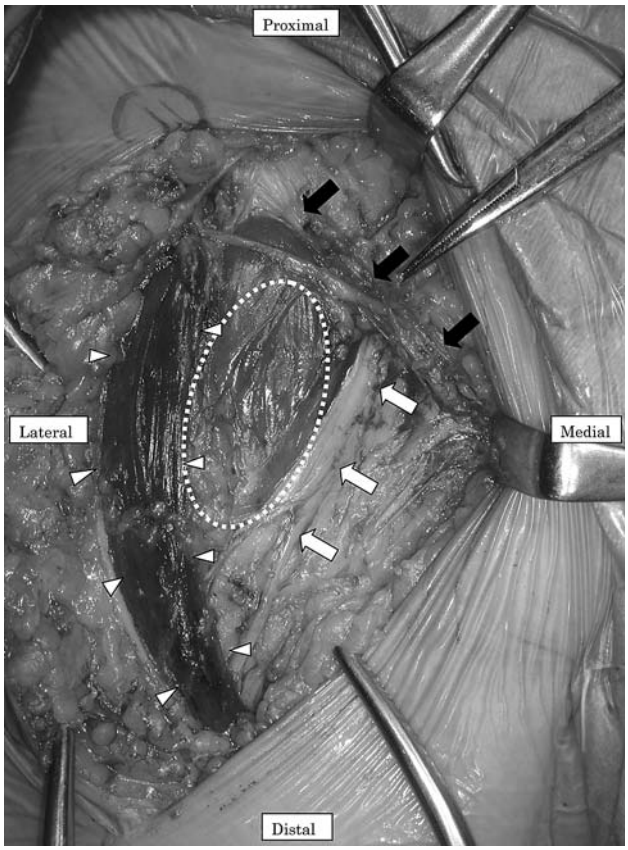

Fig. 4 The cyst was located under the iliacus muscle (within the *dotted white circle*), which was shifted medially, and the quadriceps (*white arrowhead*) was shifted laterally. The femoral nerve (*white arrow*) was pinched between the cyst and inguinal ligament (*black arrow*)

Fig. 5 Plain XP at the last examination demonstrated total hip arthroplasty and there was no sign of recurrence

complaints at the last examination 18 months after the first operation (Fig. 5).

Discussion

The causes of iliopectineal bursitis are reported to be complicated with various diseases such as rheumatoid arthritis. The iliopectineal bursa is located beneath the iliacus muscle, in the front of the capsule of the hip joint, and, inside, the femoral vessels, nerve, iliopectineal eminence, and its lateral edge reach the anterior inferior iliac spine. The iliopectineal bursa is the largest bursa in the body, and approximately 14% of the human population have a connection between it and the hip joint [9]. However, chronic inflammation of the hip joint creates a perforation between the hip joint and iliopectineal bursa.

Coventry proposed three theories regarding the cause of the enlargement of an iliopectineal cyst in RA [10]. The first theory is that overproduction of synovial fluid in a rheumatoid joint might increase the intra-articular pressure and distend the capsule of the joint at a weak point .The second theory is that the iliopectineal bursa may become involved in the rheumatoid process with the formation of excessive quantities of fluid, enlargement of the bursa, and hypertrophic and villous proliferation of the bursal lining. Necrosis from degenerative and rheumatoid change and pressure in the bursa produce a channel communicating with the hip joint. The third theory is that necrosis of a subcutaneous periarticular rheumatoid nodule could result in the formation of a juxtaarticular cyst simulating the appearance of a synovial cyst.

This patient was taken by sudden multiarticular pain, and she could not walk with difference of length of the leg.

We suppose that there is a connection for the condition of RA and the destruction of the hip joint.

We thought that the femoral head was rapidly destroyed in our case because its osteolysis progressed within a short period, and the low-level cortical atrophy means that little time has passed since she became unable to walk. This case was more consistent with the first theory because destructive change of the hip joint was so rapid and severe that the femoral head disappeared and joint synovial proliferation was marked. However, it is unclear whether the communicating channel had existed earlier or was produced by synovitis. Shiga reported that overproduced joint fluid flowed into the bursa through the channel, which had a valve-like mechanism, and the bursa became enlarged [11]. However, we could not verify a valve-like mechanism in our case.

There are many reports on enlarged iliopectineal bursitis in RA, but there are only four single reports on femoral neuropathy as far as we know. Kim reported that only two patients in a series of 119 surgically treated femoral nerve lesions at intrapelvic and thigh levels had bursitis [12], but they did not specify about RA. The prevalence of femoral neuropathy with bursitis in RA has not been clarified.

The femoral nerve has a long retroperitoneal course. It arises from the dorsal division of the ventral rami of the second, third, and fourth lumbar nerves within the psoas muscle. Passing obliquely through the muscle, it emerges from the lateral border below the iliac crest and runs down in a shallow groove between the psoas muscle and iliacus muscles to enter the thigh behind the inguinal ligament, lateral to the femoral sheath and vessels [13]. The bursa enlargement had progressed to the proximal area beneath the major psoas muscle, and passed under the inguinal ligament. Because there is a space in the pelvic cavity, the bursa had extended there. Between the expanded bursa and inguinal ligament, the femoral nerve was entrapped, leading to neuropathy [14].

On the other hand, the most common cause of spontaneous femoral nerve palsy is retroperitoneal hemorrhage. If palsy is in the early stage, the prognosis of femoral nerve palsy is good, and conservative management can be selected. However, if palsy is progressive, surgical excision of the mass should be performed [15, 16].

It was reported that steroid therapy was effective in bursae with RA without palsy [4] or with improving palsy [17], but the present case had severe palsy and so we decided on exploration. Some reports have indicated that total hip arthroplasty should be performed at the same time as cyst resection in cases without palsy. In our case, we decided to perform total hip arthroplasty for destruction of the hip joint during a subsequent surgery, because she could not control her right leg voluntarily and we thought there was a high risk of dislocation. Other additional reasons were the presence of weak skin over the cyst which might have delayed wound healing and cause infection, and the requirement of immediate chemotherapy including immunosuppression for RA after the operation to improve multiple arthritis.

Our patient fully recovered within about 6 months, symptoms were completely resolved, and there was no sign of recurrence at the last examination 18 months after follow-up.

This case represented RA developing in an older patient which worsened within a short period. Clinical control of RA was insufficient because of its rapid progress, and the cyst was huge without a valve-check mechanism, to leading to femoral nerve palsy. The iliopectineal bursa had expanded and progressed rapidly to the proximal area under the inguinal ligament, which was the cause of femoral nerve palsy. The prognosis of femoral nerve palsy after release was good.

Conflict of interest statement This article does not contribute to any conflict of interest.

References

1. Toris T, Chosa H, Kitano M. Rheumatoid synovial cyst of the hip. Clin Orthop. 1978;137:191–3.
2. Letourneau L, Dessureault M, Carette S. Rheumatoid iliopsoas brusitis presenting as unilateral femoral nerve palsy. J Rheumatol. 1991;18:462–3.
3. Kataoka M, Toris T, Nakamura M, Uchida K. Iliopsoas bursa of the rheumatoid hip joint. A case report and review of the literature. Clin Rheumatol. 1995;14:358–64.
4. Generini S, Matucci-Cerinic M. Rheumatoid synovial cyst of the hip joint. Clin Exp Rheumatol. 1993;11:549–51.
5. Warren R, Kaye JJ, Salrati EA. Arthrographic demonstration of an enlarged ipiopsoas bursa complicating osteoarthritis of the hip joint. J Bone Joint Surg. 1975;57A:413–5.
6. Kolmert L, Persson BM, Herrlin K, Ekelund L. Iliopectinael bursitis following total hip replacement. Acta Orthop Scand. 1984;55:63–5.
7. Carr CR, Berley FV, Davis WC. Pigmented villonodular synovitis of the hip joint. J Bone Joint Surg. 1954;36-A:1007–13.
8. Baito C, Nashiki H, Ishimaru A, Miyamoto M. A case report of femoral nerve palsy due to the iliopectineal bursitis in a hemodialysis (in Japanese). Seikeigeka. 1994;45:60–3.
9. Chandler SB. The iliopsoas bursa in man. Anat Rec. 1934;58:235–40.
10. Coventry MB, Polley HF, Weiner AD. Rheumatoid synovial cyst of hip. J Bone Joint Surg. 1959;41A:721–30.
11. Shiga T, Watanabe N, Sugita M, Kamada Y, Inoue S, Kubo T. Two cases of osteochondromatosis which developed in the iliopectineal bursae of an osteoarthritic hip. Mod Rheumatol. 2001;11:360–2.
12. Kim DH, Murovic JA, Tiel RL, Kline DG. Intrapelvic and thighlevel femoral nerve lesions: management and outcomes in 119 surgically treated cases. J Neurosurg. 2004;100:989–96.

13. Eustace S, Mccarthy C, O'Bryne J, Breatnach E, Fitzgerald E. Computed tomography of the retroperitoneum in patients with femoral neuropathy. Can Assoc Radiol J. 1994;45:277–83.
14. Kokubun K. Anatomical and clinical study on the femoral nerve lesion (in Japanese). Nippon Seikeigeka Gakkai Zasshi. 1983;57(1):65–77.
15. Shane P, Jeffrey C, Ronald F, Omaida V, Marc M. Femoral neuropathy following retroperitoneal hemorrhage: case series and review of the literature. Ann Vasc Surg. 2006;20(4):536–40.
16. Mori S, Tamura T, Komatsubara S, Kawaguchi Y, Todo S, Inoo M, et al. A case of femoral nerve palsy caused by iliopectineal bursitis associated with rheumatoid arthritis. Mod Rheumatol. 2004;14:274–8.
17. Matsumoto T, Juji T, Mori T. Enlarged psoas muscle and iliopsoas bursitis associated with a rapidly destructive hip in a patient with rheumatoid arthritis. Mod Rheumatol. 2006;16(1): 52–4.